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Original Communications

PRIMARY PULMONARY MALIGNANCY

FACTORS INFLUENCING SURVIVAL

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IN THE fifteen years which have elapsed since Evarts Graham performed the first successful pneumonectomy for carcinoma of the lung on a human subject, the concepts formerly held concerning this disease have been radically modified. It is now recognized that instead of being rare, it is a common type of malignancy, being preceded in frequency only by carcinoma of the gastrointestinal tract, skin, and prostate. It is now recognized that instead of being incurable, it is susceptible to cure in a significant proportion of cases, if the patient is seen while the growth is still confined to the lung. Finally, it is now recognized that resection of the malignant growth, in addition to being the only treatment from which cure can be expected, is also a perfectly practical method of therapy. A number of variations in technique have been devised, but resection has been established as the appropriate and logical form of therapy for carcinoma of the lung, and basic technical principles for its performance have been standardized. Present problems in management are concerned not so much with the type of treatment as with how to extend the benefits of surgery to more patients and thus to increase the possible salvage.

The present communication consists of an analysis of various factors which appear to influence end-results and survival of the 548 patients with primary pulmonary malignancy observed at Charity Hospital of Louisiana at New Orleans (on the Tulane University School of Medicine Service) and in private

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practice at the Ochsner Clinic from 1934, when exploration was performed on the first patient for this disease at Charity Hospital, to May 1, 1948. The same general features, apparent in previous reports,[2-6] were noted in this cumulative series, and they need be mentioned only briefly here. The incidence of primary pulmonary malignancy continues to increase, perhaps because the disease is actually more frequent, but in large part because more cases are being recognized. It occurs predominantly in men in the fifth, sixth, and seventh decades of life. No etiologic factor has been found to have special significance. With few exceptions the growths are bronchiogenic in origin. In the early stages of the disease the clinical picture is not characteristic and the usual symptoms are seldom all present in any given case. The physical findings are also not characteristic. In fact, the lack of a characteristic clinical picture, particularly in the early stages, and the general failure to appreciate the frequency of the

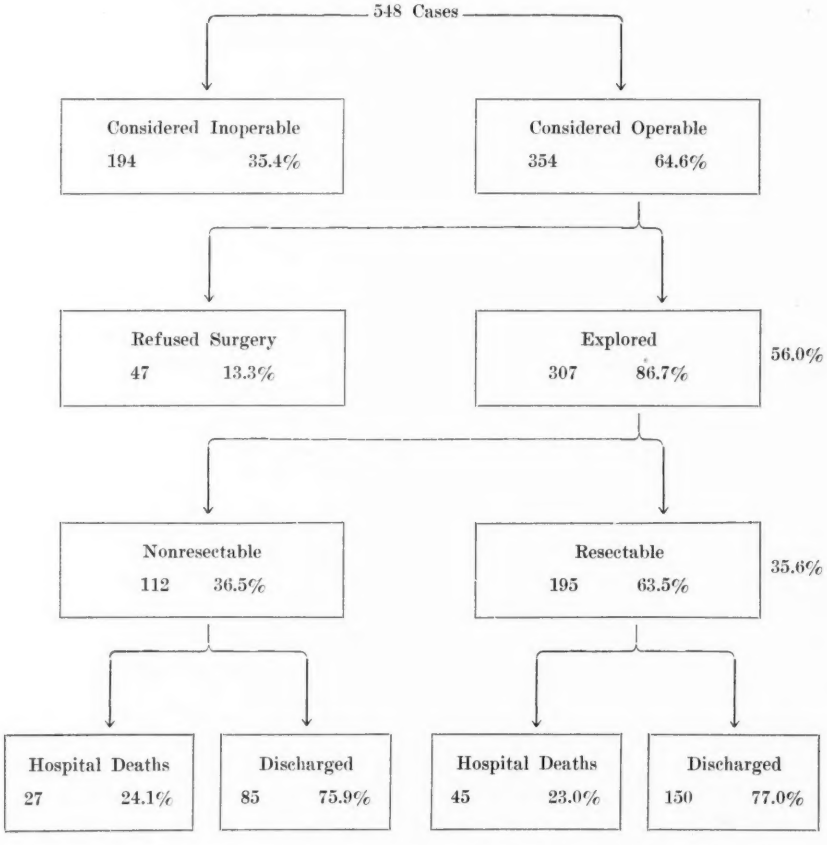

Chart 1.—Schematic representation of results in 548 cases of primary pulmonary malignancy.

disease are perhaps the two most important causes for the present low rate of cure.

It is evident that improvement is badly needed from the fact that among these 548 cases (Chart 1), the disease was far too advanced in 194 patients (35.4 per cent) to warrant even exploratory operation. Forty-seven patients considered suitable for exploration refused surgical treatment. Of the 307 patients in whom exploration was done, only 195 (63.5 per cent of the explorations and 35.6 per cent of the total series) had resectable growths. In other words, of every three patients in whom the diagnosis of carcinoma of the lung could be established on reasonable clinical grounds, one was eliminated as a candidate for operation because the growth was obviously inoperable, one had a nonresectable tumor proved at operation (or refused operation), and one had an actually resectable tumor.

These figures, however, as indicated in a previous review,[4] compare favorably with most series on record. The proportion of operability, in particular, is higher than is usually reported. This is still, however, an unsatisfactory showing, and we all share the responsibility of improving it.

DIAGNOSIS, OPERABILITY, AND RESECTABILITY

Diagnosis.—The most obvious and important method of improving the percentage of resectable cases of carcinoma of the lung—the only means now apparent by which the end results can be greatly improved—is by earlier diagnosis. It has often been stressed that the most essential factor in early diagnosis is constant awareness that the disease may be present. Primary pulmonary malignancy should be suspected as a possible diagnosis in every man over 40 years of age who has unexplained thoracic discomfort, a persistent cough, hemoptysis, or any other symptom or sign referable to the respiratory system. Nor should carcinoma of the lung be eliminated as a possibility until every diagnostic method has been exhausted, including roentgenography, bronchography, bronchoscopy, and cytologic examination of the sputum or bronchial secretions.

Roentgenography should be the initial step in the diagnostic routine in every suspected case. In seven patients without pulmonary symptoms in whom no pulmonary neoplasm was suspected, carcinoma of the lung was discovered on routine roentgenography of the chest and proved to be resectable. Such an experience clearly indicates the great value of roentgenography as part of the routine medical examination of all men in middle life and beyond.

Of all diagnostic methods bronchoscopy with biopsy is the most accurate, though unfortunately it is not applicable in all cases because of the peripheral location of some of the tumors. Of the 195 cases of resection in this series, bronchoscopy was done in 161 (82.6 per cent) and not done in the remaining cases because the lesion was located so far out in the bronchial tree that it obviously could not be visualized (Chart 2). Positive evidence of malignancy by bronchoscopic biopsy was obtained in only 37.9 per cent of the 195 cases of resection. Thus, of the entire series of resectable lesions, bronchoscopy provided a positive diagnosis in only a little more than one-third of the cases. This is

readily explained by the location of the tumors as shown by a careful pathologic examination of the resected specimens. Whereas there was some variation according to the histologic character of the growth, the lesion was situated peripheral to the lobe bronchi in approximately one-half the cases (Fig. 12). The chance of bronchoscopic visualization of the more centrally located tumors is further reduced by the fact that a large proportion of them originated in the upper lobe bronchi beyond the range of easy bronchoscopic visualization. It is obvious from these considerations that bronchoscopy has definite limitations as a diagnostic procedure and that a large proportion of patients with carcinoma of the lung would be denied proper treatment if operation were limited to those with positive results of biopsy.

Cytologic examination of the sputum or of the bronchial secretions is another important method and one which may prove, as experience with it increases, to be the most practical and most useful of all.

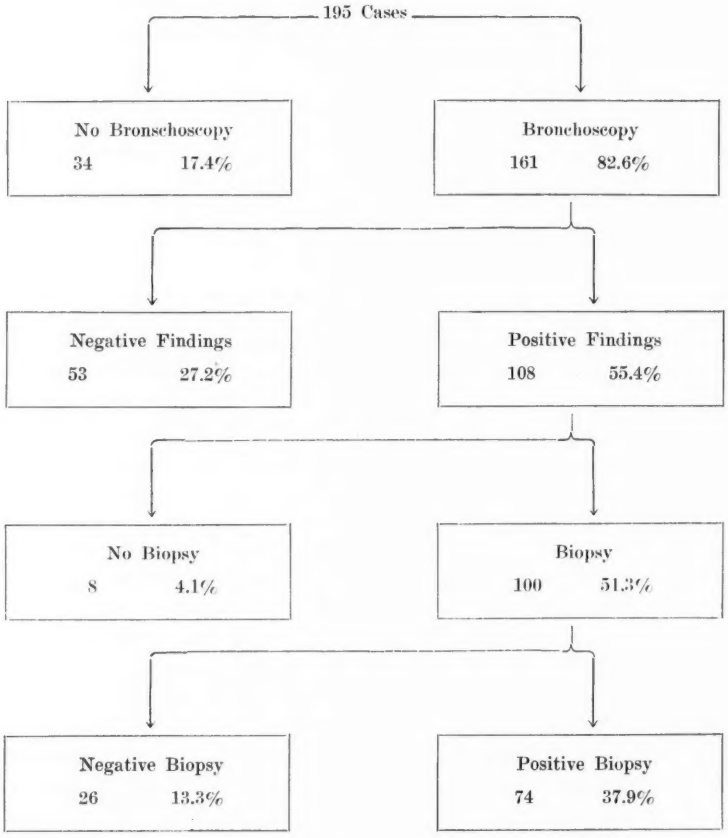

Chart 2.—Schematic representation of results of bronchoscopy in 195 cases of resection for primary pulmonary malignancy.

If a probable malignancy of the lung cannot be positively identified or excluded by any of these methods, then the surgeon is fully justified in recommending exploration of the chest. Exploratory thoracotomy is a procedure which now carries a minimum risk and if it is not applied in properly selected cases, many patients with carcinoma of the lung will be denied the opportunity for cure. A positive diagnosis was established prior to surgical exploration in 117 (60 per cent) of our 195 cases of resection and in 208 (67.8 per cent) of the entire series of 307 cases in which exploration was done.

Extension of Criteria of Operability.—Another method of increasing the proportion of resectable cases of primary pulmonary malignancy is by broadening the criteria of operability or, more accurately, by restricting the contraindications. There are few reasons, other than absolute inoperability, for not carrying out exploration on the chest of a patient with suspected carcinoma of the lung. Involvement of the phrenic or recurrent laryngeal nerves is not a definite contraindication. Such involvement was present in several patients in this series in whom resection proved possible. The definite presence of distant metastases, obvious involvement of the trachea or carina, and the presence of malignant cells in the pleural fluid indicate that the growth has passed beyond the stage of operability. In the absence of these findings it is our practice to consider operation, since exploration frequently reveals that a lesion which clinically appears to be inoperable is actually resectable.

The proportion of resectable cases in this series was further increased by the use, in addition to pneumonectomy, of certain supplemental procedures in cases in which the growth had extended beyond the substance of the lung. These procedures included resection of a portion of the pericardium with intrapericardial ligation of the hilar vessels (19 cases), suture of the auricle (10 cases), and resection of part of the diaphragm (9 cases), or thoracic wall (11 cases). Supplemental procedures were necessary only when the neoplasm had extended beyond the borders of the lung. Hence, these resections were all considered palliative and some surgeons might even question the wisdom of operation on such extensive growths. We do not share this doubt. We perform the palliative operation whenever the risk entailed is not excessive for two reasons: (1) the patient's remaining span of life is more comfortable when involved, necrotic, and infected pulmonary tissue is removed, because he is spared the sepsis and continued exudation which he would otherwise be obliged to endure. The procedure, therefore, seems warranted from the humanitarian standpoint alone. (2) There is some evidence, as will be pointed out later, that palliative resection favorably influences the period of survival. In fact, some of these patients have survived five years with no evidence of recurrence.

ANALYSIS OF RESULTS

Mortality and Causes of Death

There were 45 hospital deaths (23 per cent) in the 195 patients in whom resection was possible (Fig. 1). An analysis of the hospital death rate from the standpoint of chronology reveals a steady improvement with the passage

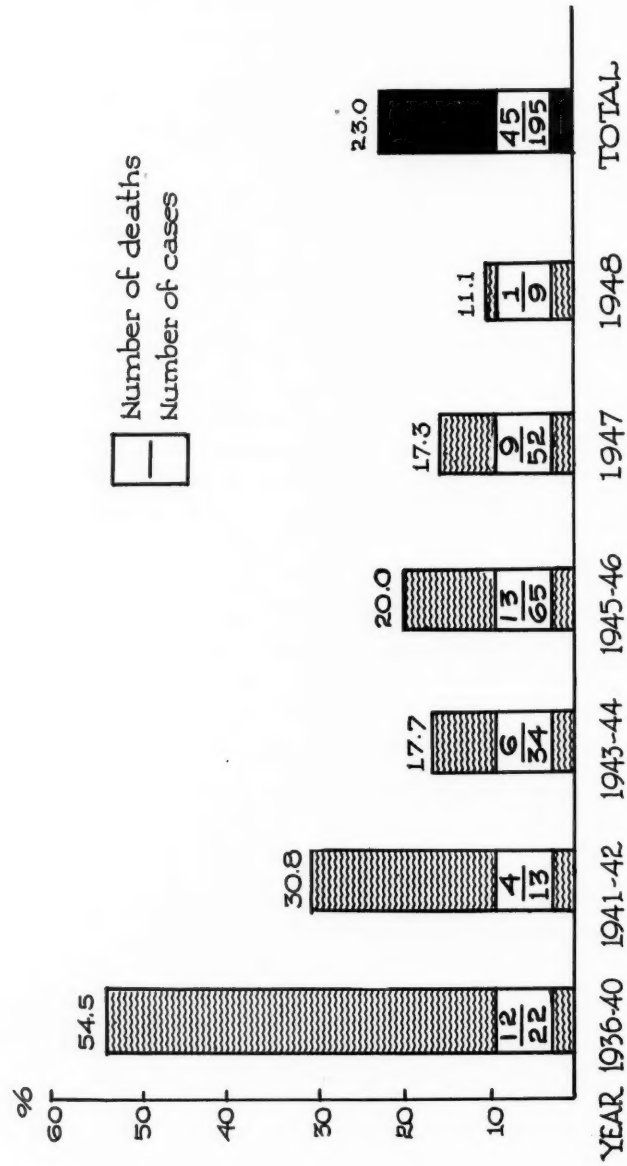

Fig. 1.—Hospital mortality in 195 cases of primary pulmonary malignancy treated by resection.

of time. The mortality for the 1947-1948 series (10 of 61 cases, 16.4 per cent) is less than one-third of the mortality rate for the first five-year period of the analysis (12 of 22 cases, 54.5 per cent). It may reasonably be assumed that the progressive reduction is a reflection of improvements in surgical technique, in anesthesia, and in preoperative and postoperative care.

The present mortality of 16.4 per cent for the 1947-1948 series is relatively high, it is true, but an analysis of the causes of death in this series (Fig. 2) leaves considerable doubt that it can be materially reduced. Such causes as hemorrhage, pneumothorax, and anesthetic complications are chiefly due to technical errors and are therefore preventable, but they accounted for only a small part of the mortality in recent cases. Possibly some deaths from postoperative empyema can be prevented, but the use of sulfonamides and penicillin does not seem to be the answer to the problem.[4, 5]

To a certain extent the major cause of death after pneumonectomy may be considered beyond the surgeon's control. In this series, as in most reported series, almost one-half of all deaths following operation were due to cardiovascular disturbances, which are frequently present, together with other constitutional diseases, in many patients in the age group in which pulmonary malignancy is most frequent. They represent an unavoidable element of risk which must be assumed in pneumonectomy for this condition unless the criteria of operability are so strictly limited that patients with cardiovascular disease are excluded from surgical treatment and are thus denied their only chance of life. In other words, the postoperative mortality of carcinoma of the lung might thus be reduced, but the final mortality of the disease would undoubtedly be increased. Perhaps with greater care in the preparation of these patients for operation the risk of cardiovascular deaths can be reduced and for this reason a cardiologist has been added to our thoracic team.

In the total series of 195 cases of resection the procedure was considered palliative in 138 (70.8 per cent) and curative in 57 (29.2 per cent), the former being determined by extension of the lesion beyond the borders of the lung either grossly or by histologic demonstration of regional lymph node metastasis and the latter by the absence of these criteria.* The fact that in 7 out of 10 cases in which resection was possible the lesion was recognized as being beyond the likelihood of surgical cure further emphasizes the need for earlier diagnosis.

The operative risk is potentially greater in palliative than in curative resections as is shown by the operative mortality in our series, the corresponding figures being 25.3 per cent and 17.5 per cent (Fig. 3). It is interesting to observe, however, that since 1942, with improved management of poor risk patients, the risk of operation in the palliative cases, which was formerly high, has been reduced until it is now about the same as that in the curative cases (Fig. 3).

*In a few cases classified as curative resections in our previous publication,[4] histologic studies of the original material revealed lymph node metastasis. These cases have now been transferred to the palliative group. For this reason some differences may be observed between the results here presented and those previously published.

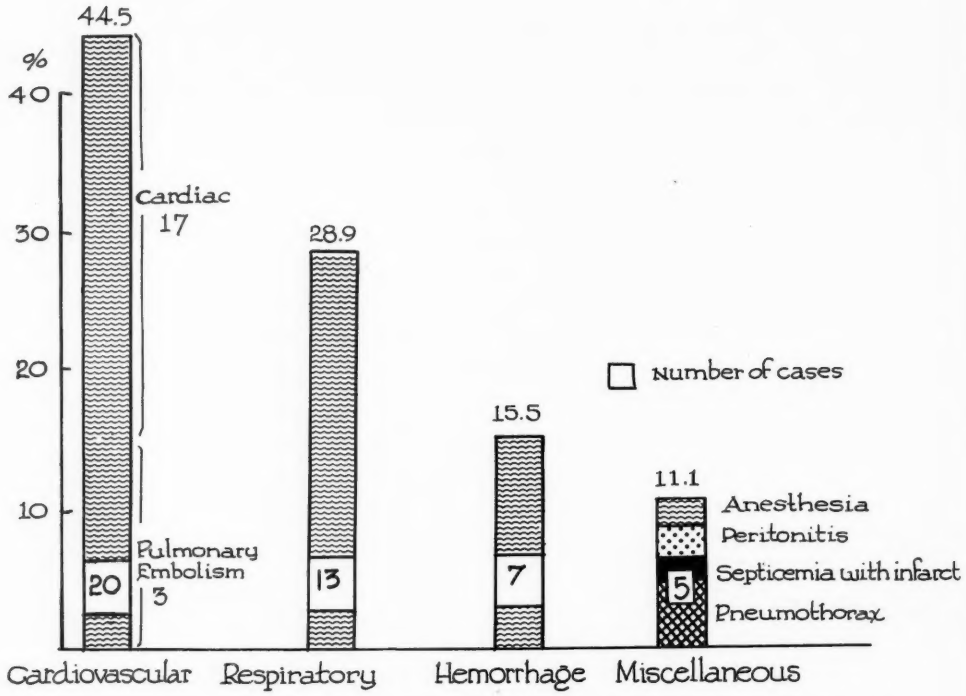

Fig. 2.—Causes of death in the 45 patients dying in the hospital among the 195 patients with primary pulmonary malignancy treated by resection.

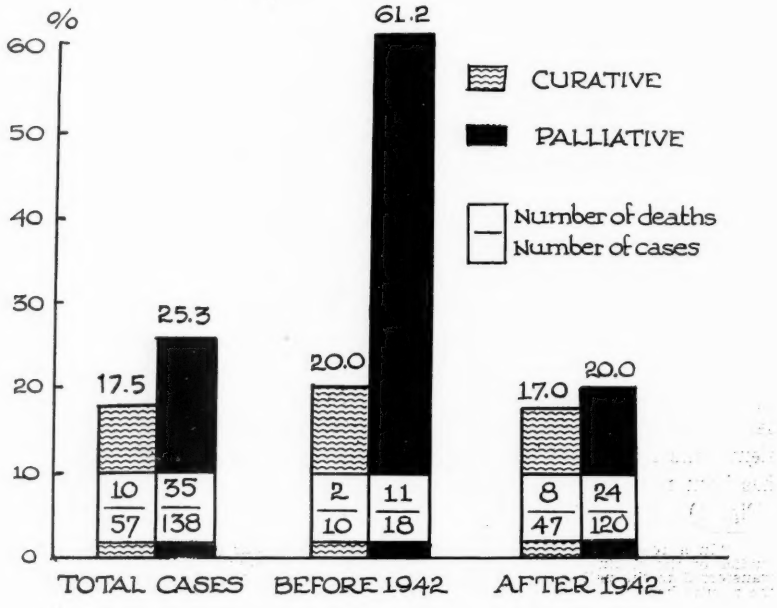

Fig. 3.—Hospital mortality in palliative and curative cases of resection.

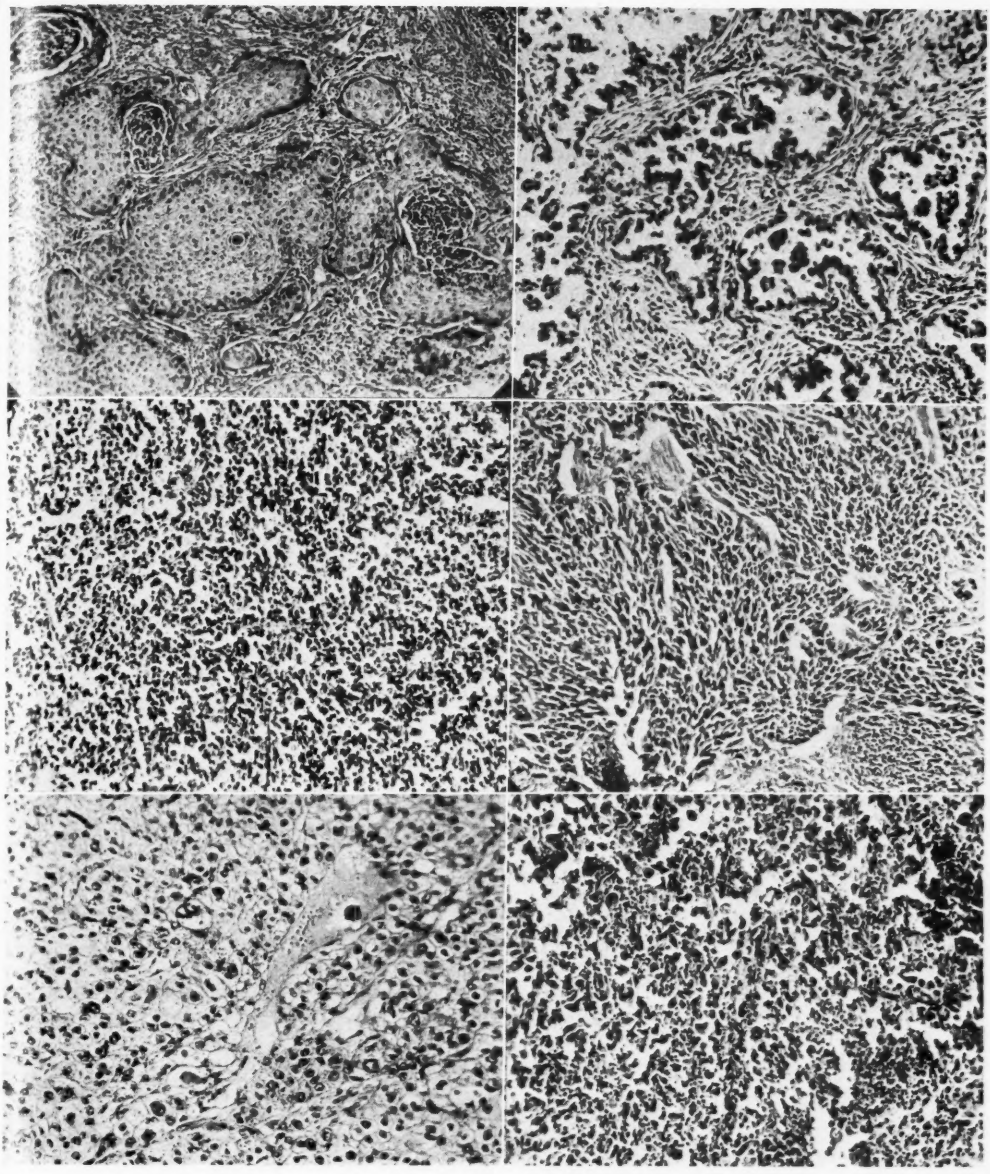

Fig. 4.—Histologic types of carcinoma of the lung. *A*, Epidermoid (squamous cell) carcinoma grade II; a few small epithelial pearls are present. *B*, Adenocarcinoma; low columnar and cuboidal tumor cells are forming irregular glandlike spaces in the fibrous stroma; small papillary projections of the tumor extend into the lumens of these spaces. *C*, Undifferentiated carcinoma, small round-cell type; the tumor is made up of fairly uniform small, round, dark-staining cells without any distinctive arrangement. This tumor can be confused with lymphosarcoma unless careful attention is paid to the structure of the nuclei and to the occasional regions in which the tumor cells are arranged along the borders of the delicately branching fibrous stroma. *D*, Undifferentiated carcinoma, oat-cell type; this tumor is not always classed among the undifferentiated carcinomas since the characteristic fusiform shape of the cells give it a distinctive appearance. In the past such tumors have sometimes been classified as sarcomas, but study of numerous sections will usually reveal some transitions to definite epithelial structures. *E*, Undifferentiated carcinoma, large clear-cell type; this tumor may be confused with reticulum-cell sarcoma but careful study will usually show a few attempts to form glandlike structures or some small nests of keratinized cells. *F*, Undifferentiated carcinoma, unclassified; this slide shows a tumor from the heterogenous group of undifferentiated tumors which were not subclassified and which appear in the figures as "undifferentiated carcinoma, other." The tumor cells are pleomorphic and bizarre giant forms are present. The picture is complicated by the presence of inflammatory cells resulting from secondary infection.

Histologic Studies.—In order to evaluate the factors which affect survival it is important to have as complete data as possible on all aspects of each case. For this reason, the subsequent analysis has been limited to the 195 cases in which resection was done. Every patient in this series has been traced to May 1, 1948. The hospital record of each patient was reviewed, the clinical signs and symptoms were tabulated, and the precise location of the neoplasm was determined as exactly as possible from the original gross pathologic descriptions and bronchoscopic roports.

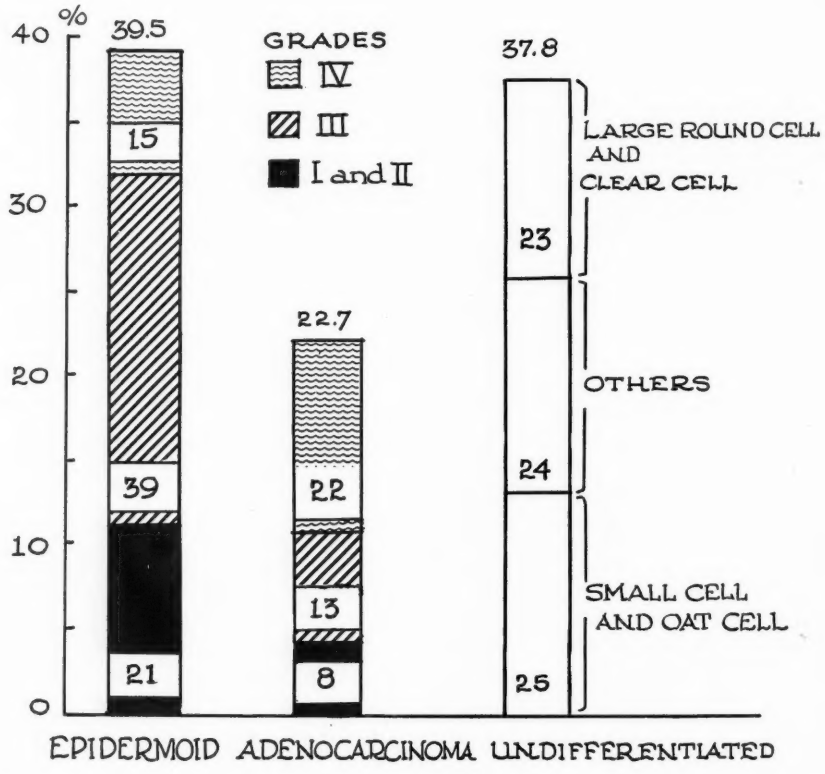

Fig. 5.—Graphic representation of distribution by histologic type and grade of the 190 cases of resection for carcinoma of the lung.

The original slides were obtained in all but one of these cases and in order to assure an objective analysis the slides were independently rediagnosed, classified, and graded by one of us (C. E. D.) without any knowledge of the original diagnosis or the clinical history or fate of the patient. Many classifications of carcinoma of the lung have been proposed, but for the purposes of this study only three major groupings were used: epidermoid (squamous cell) carcinoma, adenocarcinoma, and undifferentiated carcinoma. The cases of

epidermoid and adenocarcinoma were each further divided into four grades of histologic malignancy and the cases of undifferentiated carcinoma were arbitrarily divided into three subgroups, according to cell type: first, the small round-cell and the oat-cell carcinomas; second, the large round-cell and clear-cell tumors; and third, a miscellaneous group of irregular cell types designated as "other" (Figs. 4 and 5). All carcinomas were assigned to one of these groups and subgroups at the time the slides were reviewed without knowledge of the fate of any of the individual patients and no subsequent changes were made in this classification. Carcinoma was found in the sections from 190 of the 195 resected lungs. The lesions in the remaining 5 patients were: one low-grade neurofibrosarcoma, one Hodgkin's disease, one lymphosarcoma, one malignant melanoma, and one carcinoma diagnosed at the time of operation, but excluded from this study since the original slides were not available. Two cases originally diagnosed as fibrosarcoma and so reported in previous publications[4, 5] were reclassified as undifferentiated carcinomas and one of the cases previously reported as lymphoblastoma was reclassified as Hodgkin's disease. It is worth mentioning that each patient in whom the diagnosis is now neurofibrosarcoma, lymphosarcoma, and Hodgkin's disease is still living 12 years, 6 years, 9 months, and 3 years, 8 months, respectively, after pneumonectomy. The patient with the malignant melanoma died 2 weeks postoperatively. The patient in whom the original diagnosis was carcinoma but who was excluded from this study because the original slides are no longer available is still living 8 years, 3 months after operation.

Since the various factors affecting survival are generally evaluated in terms of the number of patients alive at yearly intervals after treatment, correlations of this character were made whenever possible. In other instances a "fatality rate" was determined by adding up the total number of years of observation on all patients living and dead in a particular group and dividing the total number of deaths by this figure to give a fatality rate in terms of the number of deaths per patient-year of observation. This is an accepted method of statistical treatment and has the advantages of taking into account the length of survival of those patients who died and at the same time including living patients of both long and short survival.

Survival Rates

Every patient in this series who survived operation and who was submitted to surgical treatment six months ago or longer has been followed up to date. Because the patients involved in these calculations were operated upon at varying times in the past, the number of cases in each category is different, and the numbers become progressively smaller as the possible survival time increases. Thus, patients operated upon only one year ago have had no opportunity to survive longer than one year. Accordingly, the survival rate in this analysis is calculated on the basis of the possible period of survival following operation. In some instances, as will be appropriately indicated, the survival rate analyses are made for the entire series of 195 cases of resection of primary pulmonary

malignancy, whereas in others it is based only upon the 190 cases of resection of carcinoma of the lung.

Of the 181 patients with primary pulmonary malignancy operated upon six months ago or longer, 99, a little more than one-half, were alive at the end of this period; the 62 deaths include the fatalities which occurred in the hospital immediately after operation (Fig. 6). At the end of one year not quite 40 per cent of the patients submitted to resection were still alive. At the end of the second, third, fourth, and fifth years the proportion of survivals was about the same for each year, ranging from 20 to 27.5 per cent. To express it differently, the survival curve drops rather rapidly within the first year after pneumonectomy for primary pulmonary malignancy, but by the second year it begins to be stabilized, and it continues almost as a plateau from the third to the fifth year. The assumption, therefore, seems warranted that a patient who lives through the second year after operation has a good chance of being alive at the end of five years.

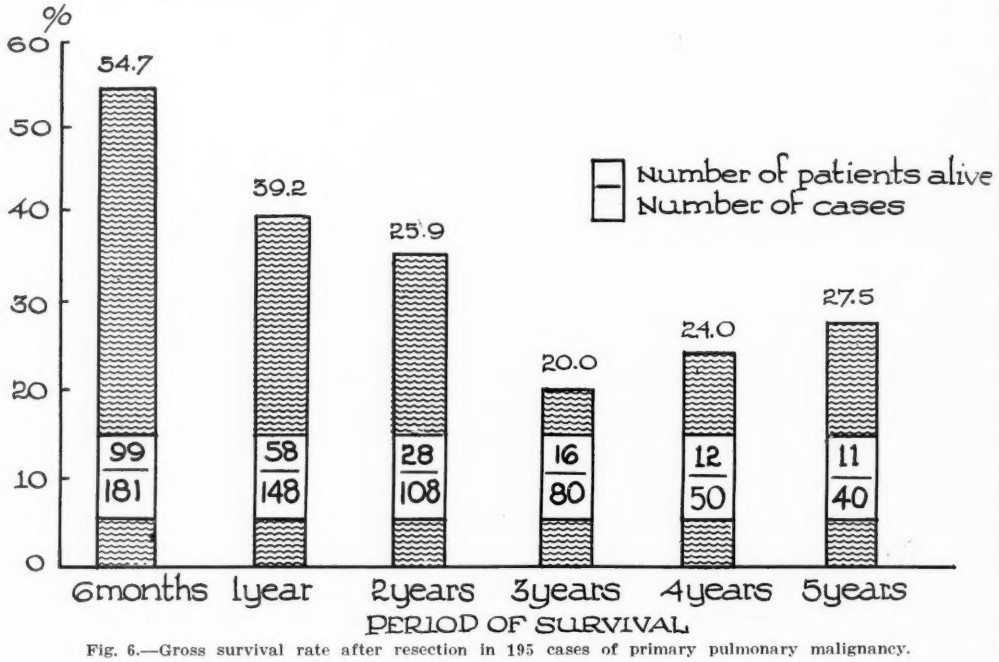

Fig. 6.—Gross survival rate after resection in 195 cases of primary pulmonary malignancy.

With the progressive improvement, through increasing experience, which has taken place in operability and in hospital mortality in this series, it was thought possible that a similar improvement might be reflected in the survival rates for the later periods of this experience. Accordingly, the patients were divided into two groups, depending upon whether the operation was carried out before or after 1942, and their survival rates were compared. No significant

difference in the rates was evident, which seems to indicate that though improvements in technical management have resulted in a reduction of the hospital mortality, they have had little if any influence on the length of survival.

Of all factors influencing the survival rate in carcinoma of the lung the most obvious is the extent of the growth, that is, whether or not it has extended beyond the borders of the lung at the time of operation. As previously stated, resections of lesions which had extended beyond the borders of the lung were considered palliative rather than curative. When the cases in this series are compared from this standpoint, the difference in survival time is, as might be expected, significantly in favor of patients who had no extrapulmonary extension of the growth at the time of operation (Fig. 7).

The question may fairly be raised as to whether patients with extension of the growth beyond the lung might not have survived just as long without operation. To determine this point, the cases in which resection was not done were divided into two categories, those in which there was no surgical treatment at all because the patient refused operation or the growths were inoperable and those in which exploration showed the growth to be inoperable, and the survival times were compared. It is realized that these cases are not strictly comparable to the cases of resection since the former are more advanced than the latter. However, the cases without resection include 47 patients in whom the lesion was considered operable but who refused operation, in two-thirds of whom the lesion probably would have been resectable. The patients in whom resection was not done showed practically the same rate of survival whether or not exploration had been carried out (Fig. 8). At the end of six months, only about 15 per cent were still alive, and at the end of three years none were alive. A comparison of the results in these cases with the results in the cases of resection shows that whereas in the former group none lived three years, among the latter 20 per cent survived this period (Fig. 9). It is also significant that 10 per cent of patients with palliative resections survived more than three years, whereas none with lesions considered inoperable lived so long. This lends support to our conviction that palliative resections are justified both for symptomatic relief and prolongation of life. A graphic comparison of the results in cases of nonresection, in all cases of resection, and in cases of resection categorized as to whether the growth was localized or had extended beyond the pulmonary tissue makes clear these differences in the survival rates (Fig. 10).

At the present time the gross survival rate in carcinoma of the lung at the end of five years is 7.7 per cent. This is not high, but it is as high or higher than the present survival rate in carcinoma of the stomach which, according to the representative literature and our own experience, does not exceed 6 to 8 per cent at the most and in some series is as low as 2 per cent. The percentage of operability in carcinoma of the stomach, furthermore, is 57 per cent, whereas that of carcinoma of the lung is 67 per cent, and the percentage of resectability in carcinoma of the stomach is 25 per cent, against 35.2 per cent for carcinoma of the lung. It, therefore, appears that the operability and rate of cure in

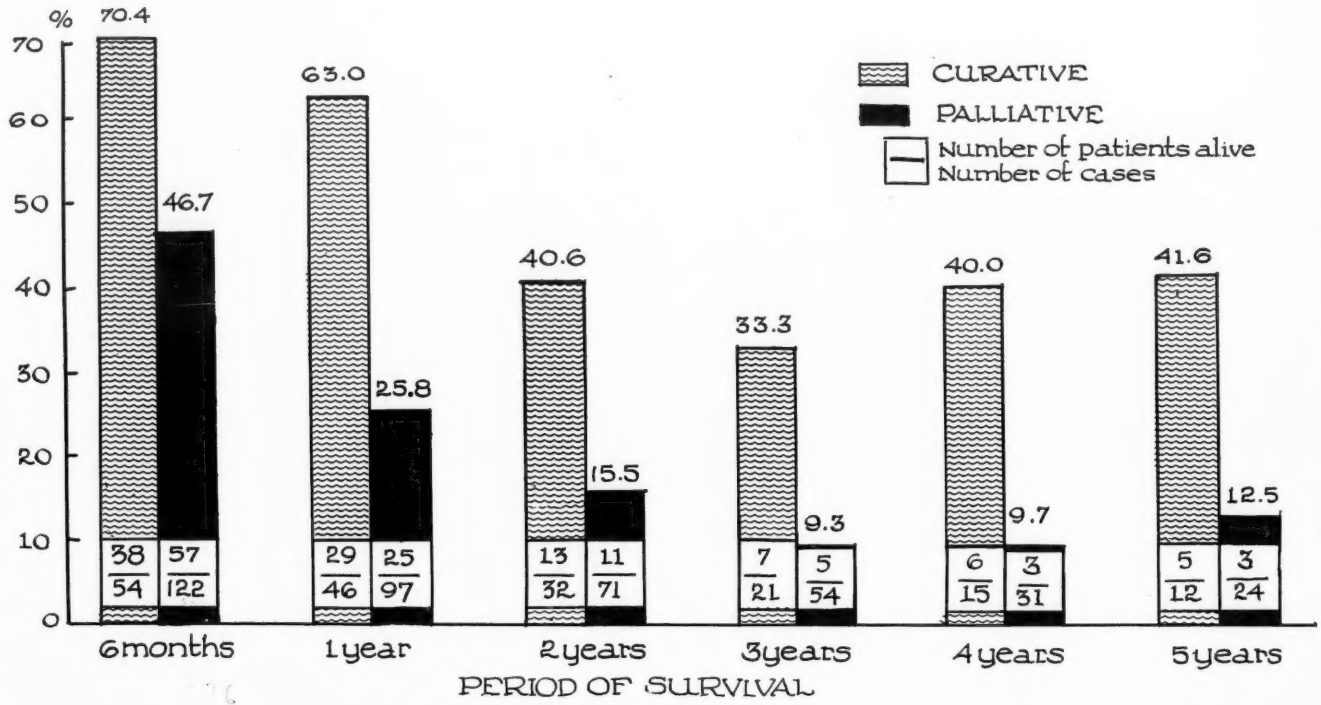

Fig. 7.—Graphic representation of survival rates after palliative and curative resection in the 190 cases of carcinoma of the lung.

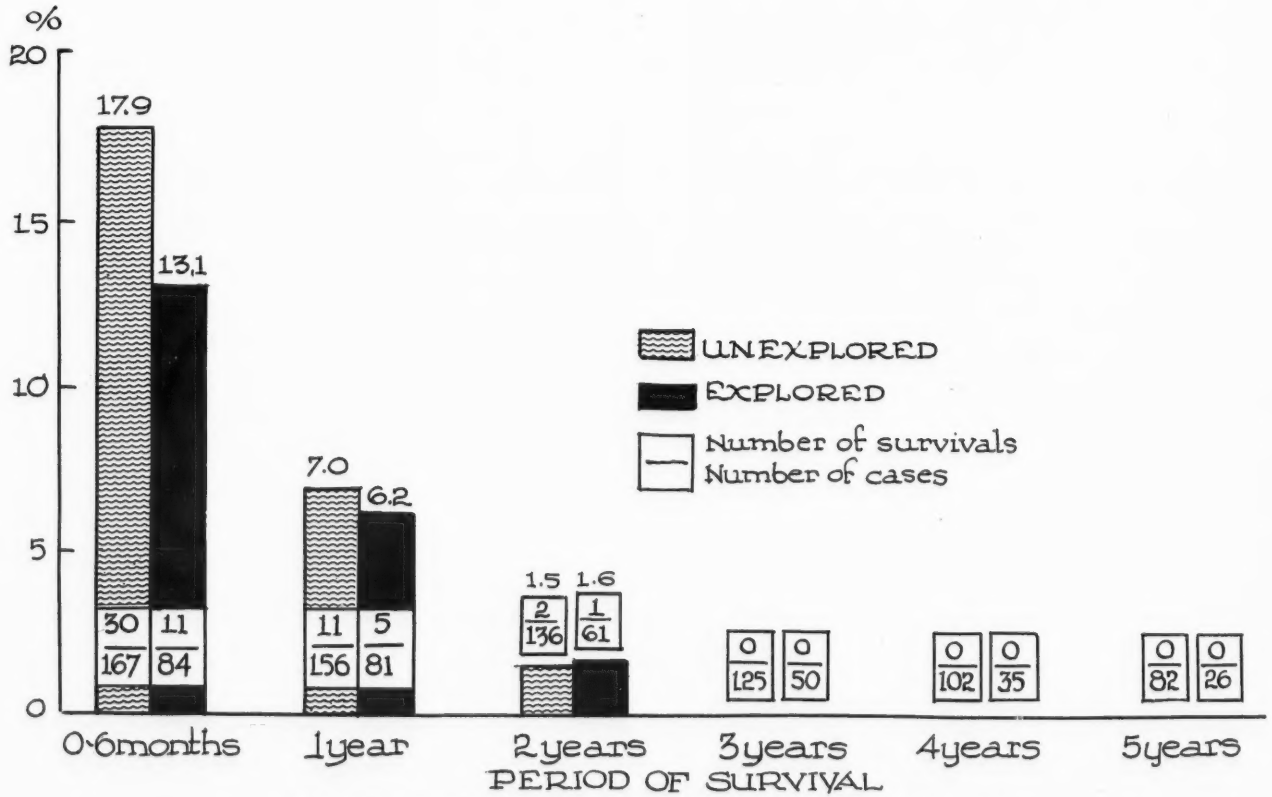

Fig. 8.—Gross survival rate in cases of nonresection for carcinoma of the lung.

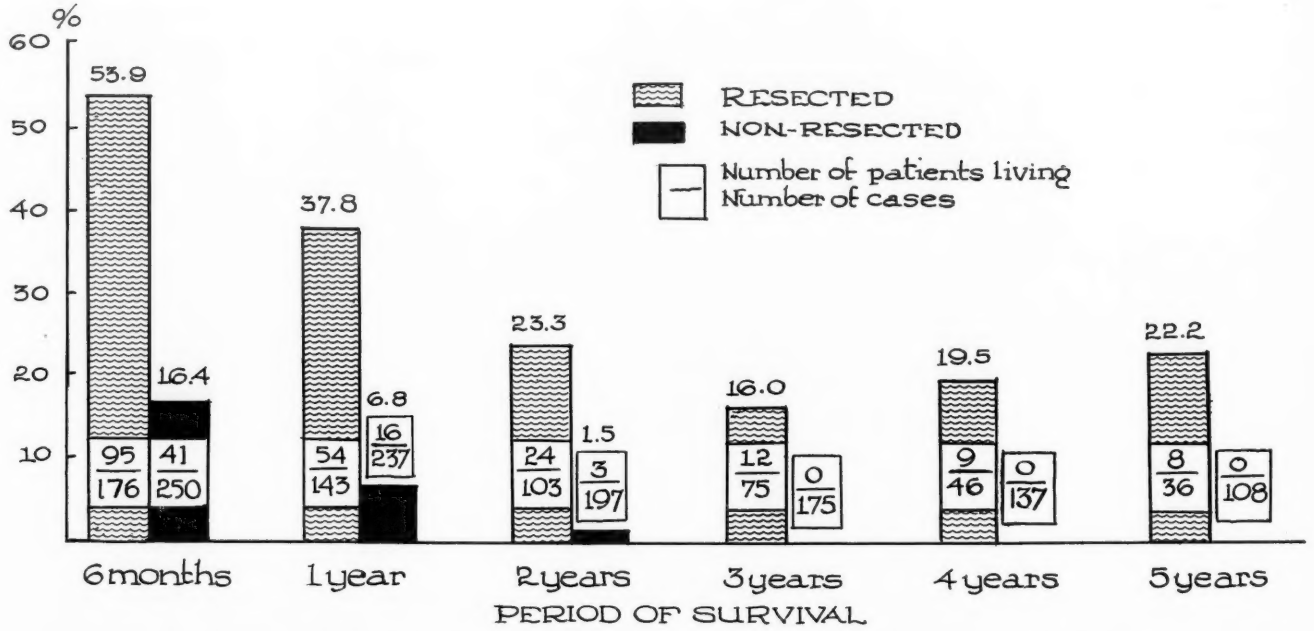

Fig. 9.—Comparison of survival rates among cases of resection and nonresection for carcinoma of the lung. The difference between the figures shown here for the cases of resection and those shown in Fig. 6 is explained by the fact that there is included in the latter the four cases diagnosed each as neurofibrosarcoma, Hodgkin's disease, lymphosarcoma, malignant melanoma, and the one case of carcinoma in which the original slides are no longer available.

carcinoma of the lung are as good as, or better than, in carcinoma of the stomach in spite of the far greater general awareness of the latter. It is probable that the results in carcinoma of the lung will be greatly improved as the problem becomes more widely recognized.

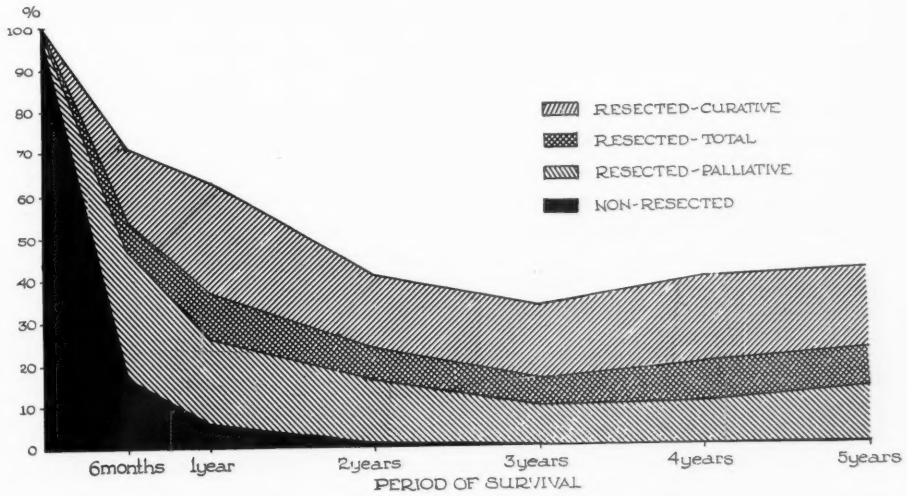

Fig. 10.—Graphic representation of survival rates in cases of resection and nonresection for carcinoma of the lung.

Special Factors Affecting Survival

Location and Extent of Tumor.—Because of the clinical impression indicated previously,[2] that greater hazards were associated with right than with left-sided lesions, an analysis was made of the fatality rate according to lobe involvement (Fig. 11). This analysis confirmed the clinical impression that right-sided lesions carried a higher fatality than left-sided lesions, the corresponding figures being 93 and 65 per cent. Among the curative cases the fatality rate was about the same for all lobes, but among the palliative cases lesions in the right upper lobe carried a much more grave prognosis than in the left upper lobe. The fatality rates for lesions arising in the main stem bronchi, lobe bronchi, and peripheral portions of the lung were about the same, the figures being, respectively, 79, 71, and 68 per cent. In the entire group central and peripheral lesions were about equally divided. The epidermoid carcinomas were predominantly central and the adenocarcinomas were predominantly peripheral (Fig. 12).

Age.—Since age is generally recognized as an important consideration in determining both the risk of operation and the survival period after operation, an analysis of this factor was made in several ways. A comparison of the age distribution by histologic types showed that a higher proportion of the epidermoid and undifferentiated forms of bronchiogenic carcinoma occur in the older

age groups than do the cases of adenocarcinoma (Fig. 13). This is more strikingly demonstrated by determining the relative incidence of each of these histologic types of carcinoma in different age periods. From this analysis it would seem that before the age of 50 years the chances of having any one of these types are about equal, but after the age of 60 years there is a two and one-half times greater chance of having an epidermoid carcinoma than an adenocarcinoma, whereas the proportion of undifferentiated neoplasms shows little change (Fig. 14).

The influence of age upon survival was determined by computing the fatality rates according to the age of the patient at the time of operation. As might be expected, the fatality rises with increasing age (Fig. 15). Since it is conceivable that this may be due in great measure to the greater operative hazard in elderly patients, a similar analysis was made excluding all deaths

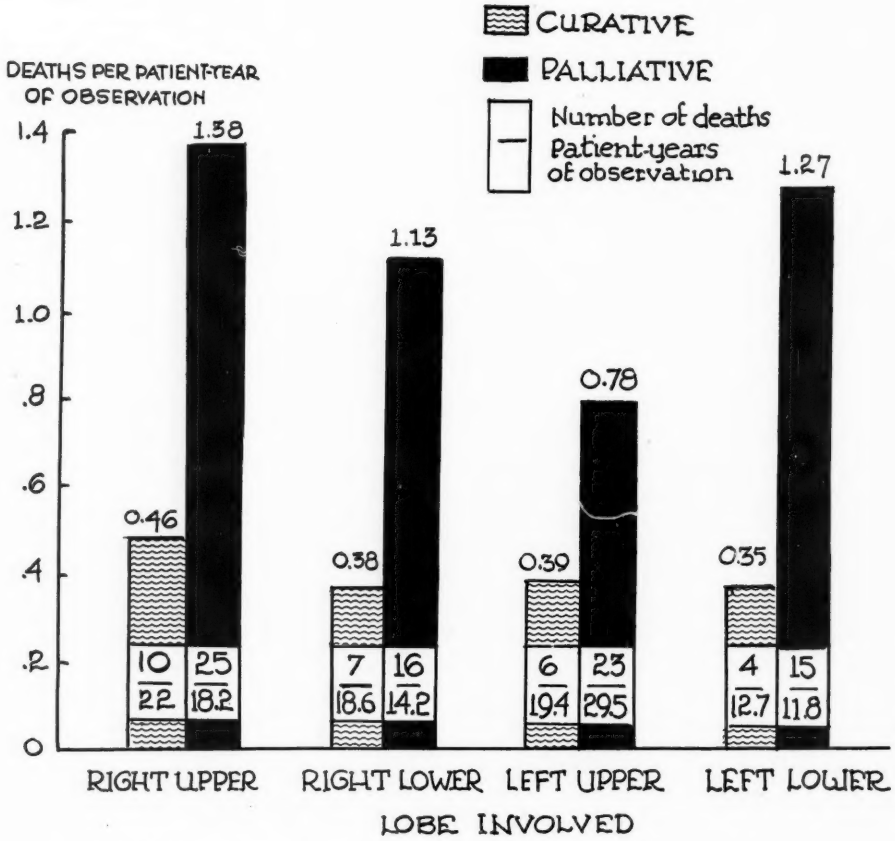

Fig. 11.—Fatality rate according to lobe involved in the 190 cases of resection for carcinoma of the lung.

occurring within thirty days after the operation. This showed that the pronounced increase in fatality after the age of 60 was due principally to early postoperative deaths (Fig. 15). This would tend to support the clinical impression that the hazards of operation become much greater with increasing age.

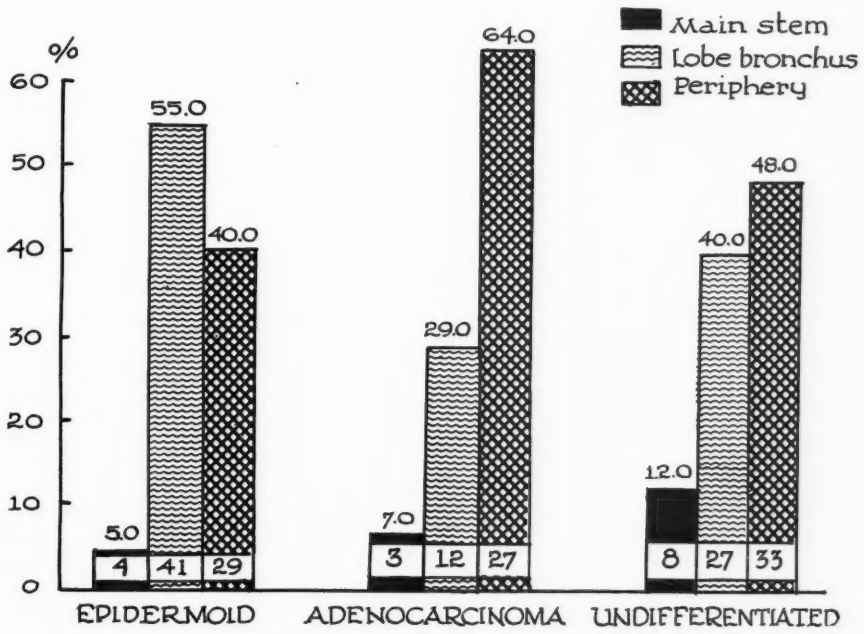

Fig. 12.—Distribution by location of lesion in the 190 cases of resection for carcinoma of the lung. The figures within the columns represent the number of cases and those above the columns represent percentage. Lesions designated as being in the "periphery" include all those originating distal to the primary bronchi.

Sex.—It has long been recognized that carcinoma of the lung is predominantly a disease of men, though no satisfactory explanation for this curious phenomenon has been advanced. In this series of 190 cases there were only 27 women (Fig. 16). Another interesting observation became apparent upon analysis of the sex distribution of our cases by histologic types. It was found that in men epidermoid carcinoma occurred more than two and one-half times more frequently than adenocarcinoma, but in women this ratio was completely reversed (Fig. 17).

Because of these differences in the relative incidence and types of carcinoma of the lung in the two sexes, it was considered desirable to determine the survival rates according to sex. The women showed a consistently more favorable prognosis over a five-year period than the men (Fig. 18). This may be explained in part by the fact that the average age of the women with resectable carcinoma of the lung was four years less than that of the men.

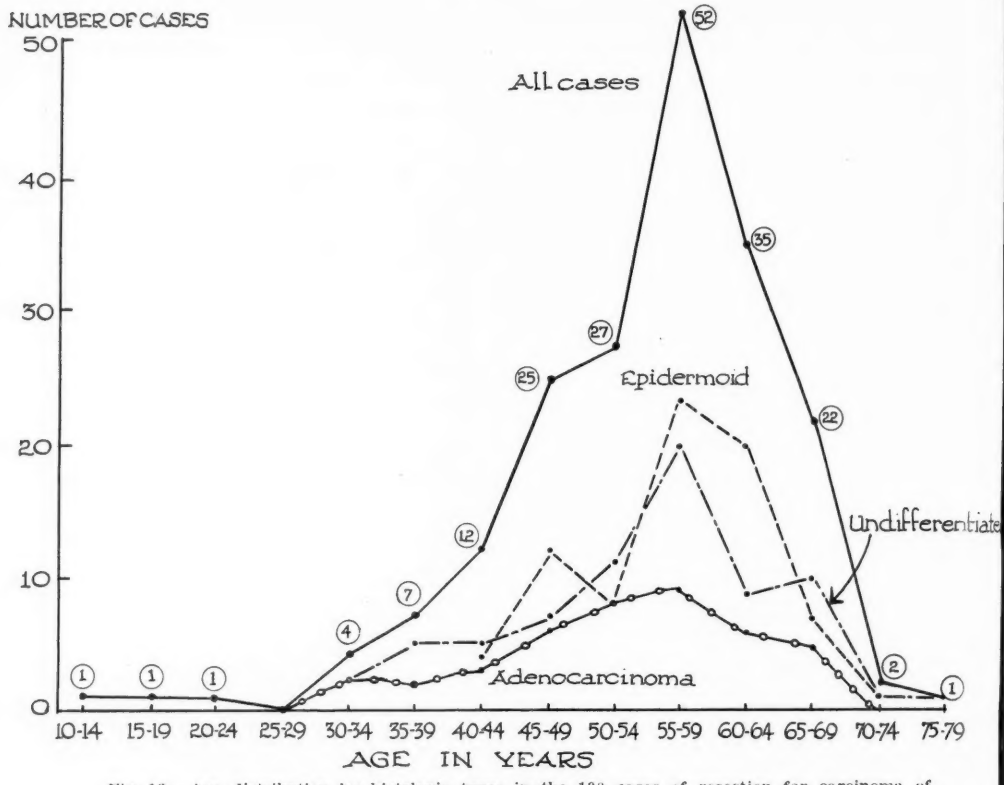

Fig. 13.—Age distribution by histologic types in the 190 cases of resection for carcinoma of the lung.

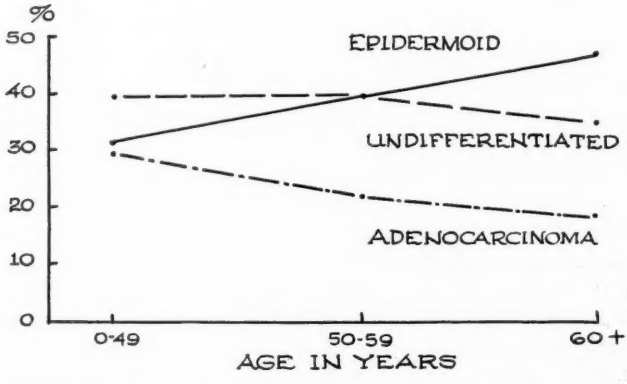

Fig. 14.—Incidence of histologic type according to age in the 190 cases of resection for carcinoma of the lung.

Survival by Histologic Types and Grades

The gross survival rate for the entire series of patients has already been presented. The patients were then studied in groups according to the histologic type of the tumors and the survival rate was computed for each group. The patients with epidermoid carcinoma showed the highest survival rate, undifferentiated carcinomas the lowest, whereas those with adenocarcinomas were intermediate (Fig. 19). Fatality rates were then computed according to the histologic grade of malignancy in each of the major histologic groups. Among

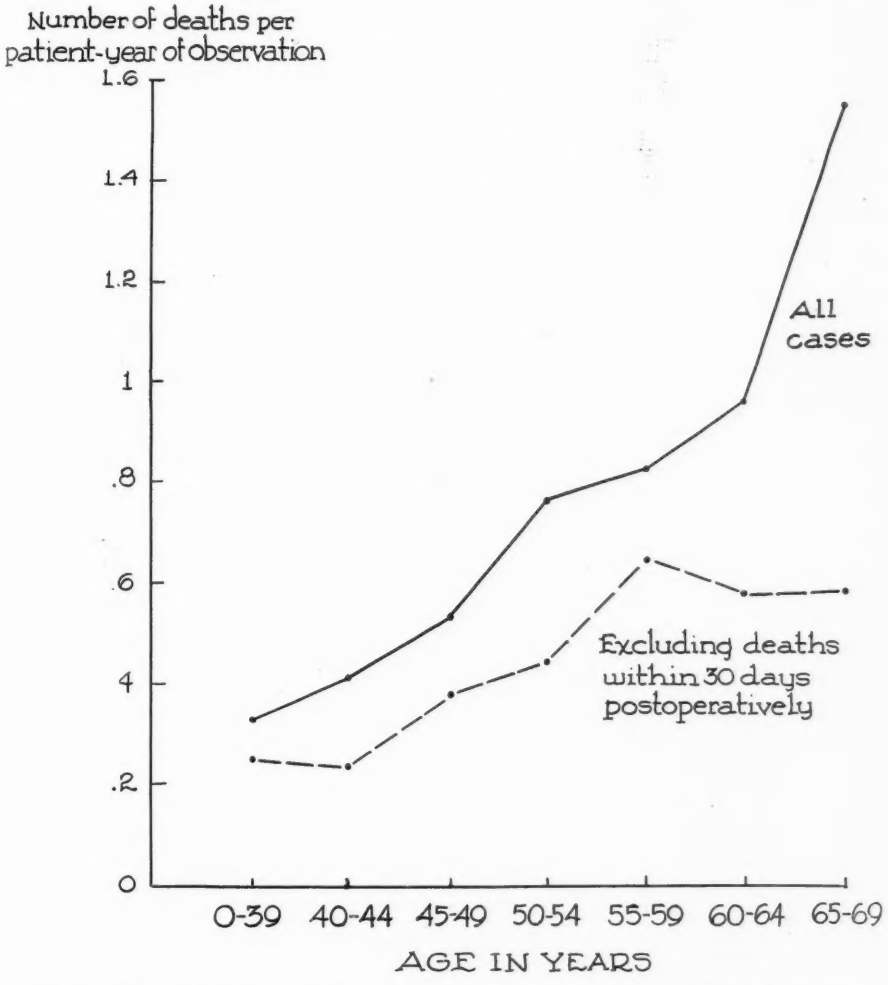

Fig. 15.—Fatality rates according to age at the time of operation in the 190 cases of resection for carcinoma of the lung.

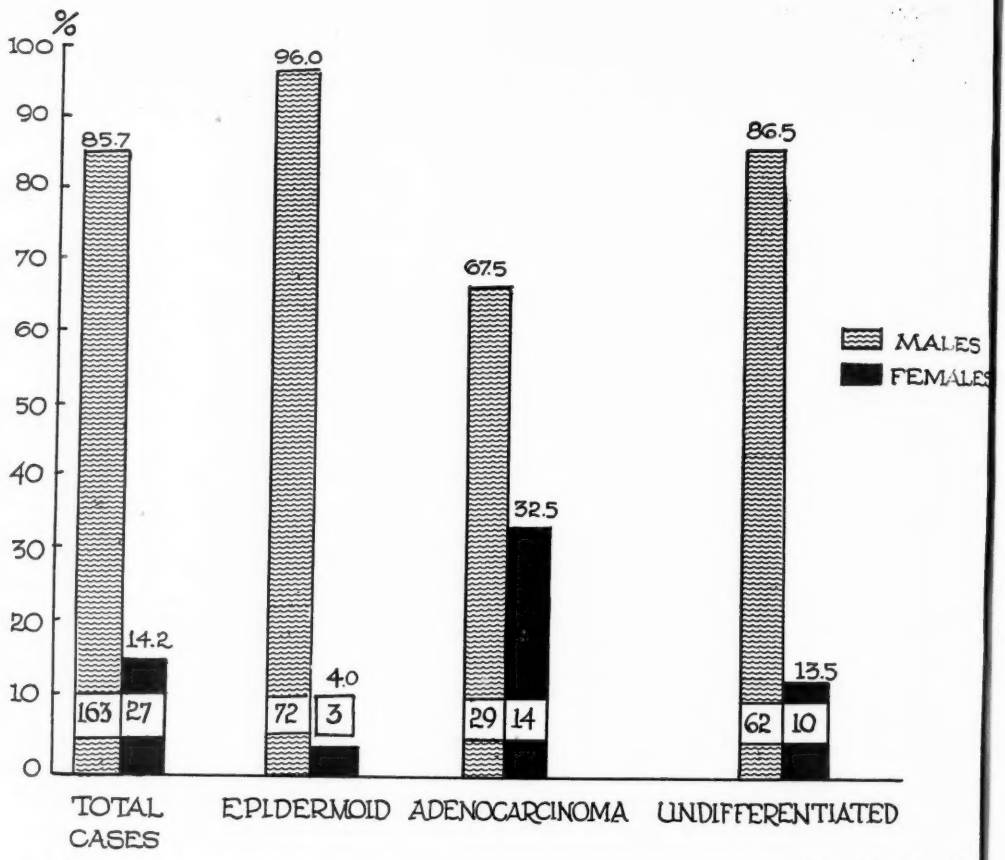

Fig. 16.—Sex distribution by histologic type in the 190 cases of resection for carcinoma of the lung.

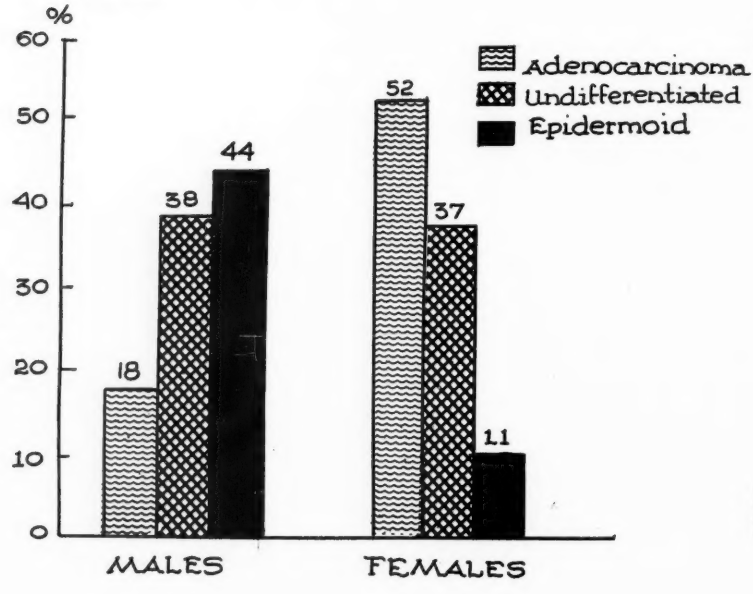

Fig. 17.—Sex distribution by histologic type in the 190 cases of resection for carcinoma of the lung.

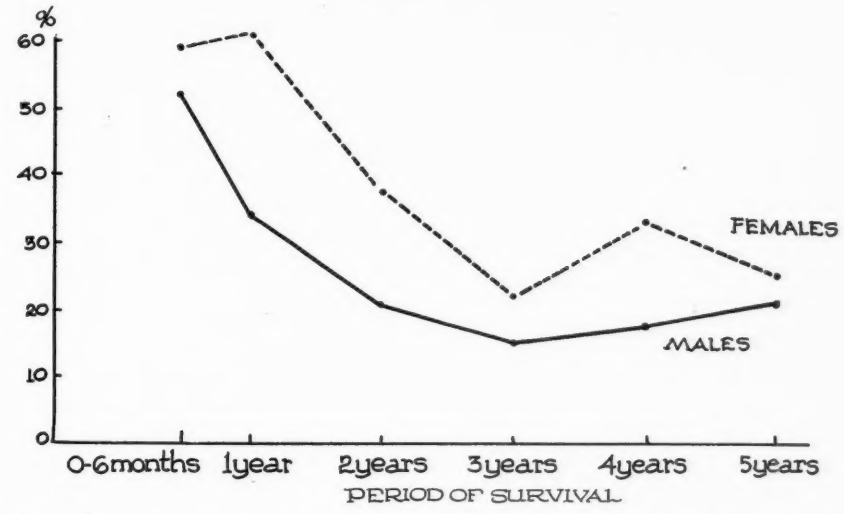

Fig. 18.—Survival rate according to sex in the 190 cases of resection for carcinoma of the lung.

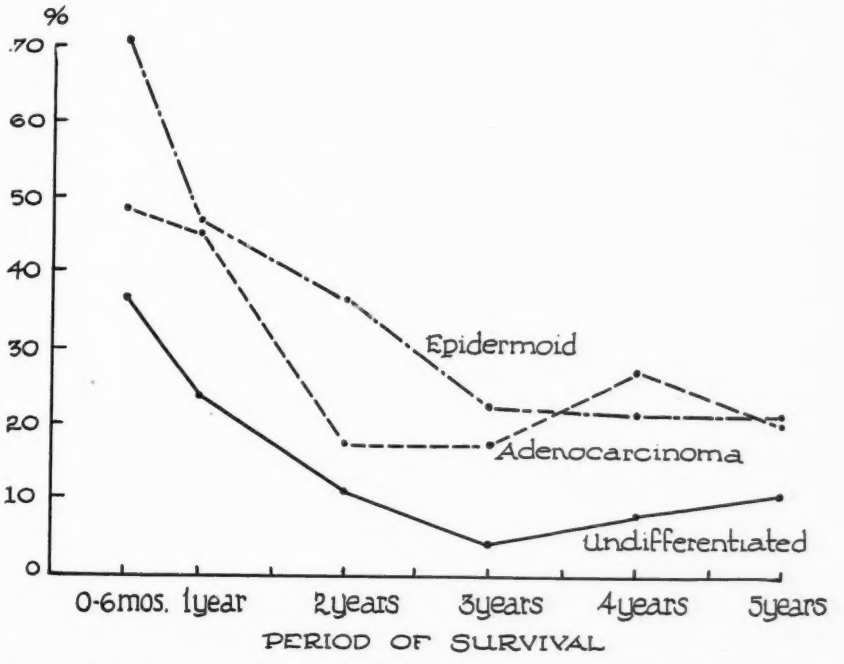

Fig. 19.—Survival rate according to histologic type in the 190 cases of resection for carcinoma of the lung.

the epidermoid carcinomas there was a progressive rise in the fatality rate from Grade I and II to Grade IV, which was apparent both in the palliative and the curative cases (Fig. 20). The same pattern was apparent among the different grades of adenocarcinoma (Fig. 21). Although the undifferentiated carcinomas were not graded, the small-cell and oat-cell tumors carried the lowest fatality, the miscellaneous group was next, and the large round- and clear-cell tumors gave by far the highest fatality rate of any group studied (Fig. 22).

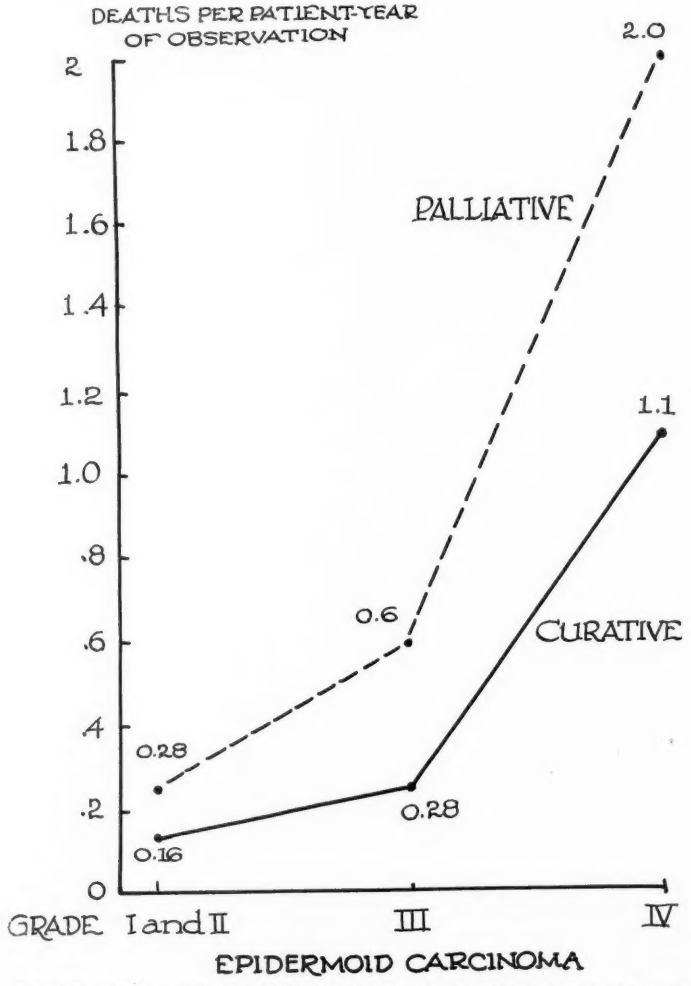

Fig. 20.—Fatality rate according to histologic grade in the 51 palliative and 24 curative resections of epidermoid carcinoma of the lung.

SUMMARY AND CONCLUSIONS

1. Among 548 patients with primary pulmonary malignancy, 194 (35.4 per cent) had inoperable lesions, 47 (6 per cent) refused operation, and 307 (56.1 per cent) had exploration. Of the 307 explorations, the lesion was found to be resectable in 195 (35.6 per cent of the total series). In brief, this means that of every three patients with carcinoma of the lung one had an obviously inoperable lesion, one had a lesion found to be inoperable at exploration (or refused operation), and one had a resectable lesion.

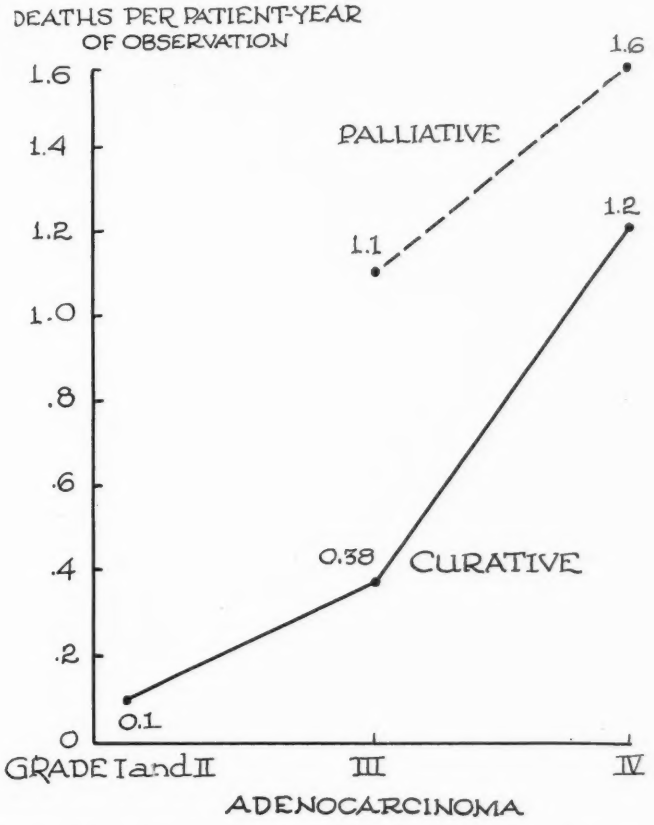

Fig. 21.—Fatality rate according to histologic grade in the 31 palliative and 12 curative resections of adenocarcinoma of the lung.

2. Positive results of bronchoscopic biopsy were obtained in only 37.9 per cent of the 195 cases of resection. In approximately the same proportion of cases, the lesions in the resected specimens were found to be within range of bronchoscopic visualization. This illustrates the diagnostic limitations of bronchoscopy and emphasizes the importance of exploration when all diagnostic methods yield only presumptive evidence of carcinoma.

3. Among the 195 cases of resection there were 45 hospital deaths. There has been a progressive reduction in hospital mortality during the period of this study from an initial figure of 54.5 per cent to 16.4 per cent. Approximately one-half of the hospital deaths were attributable to cardiovascular disturbances, one-quarter to respiratory disorders, and the remainder to hemorrhage and miscellaneous causes.

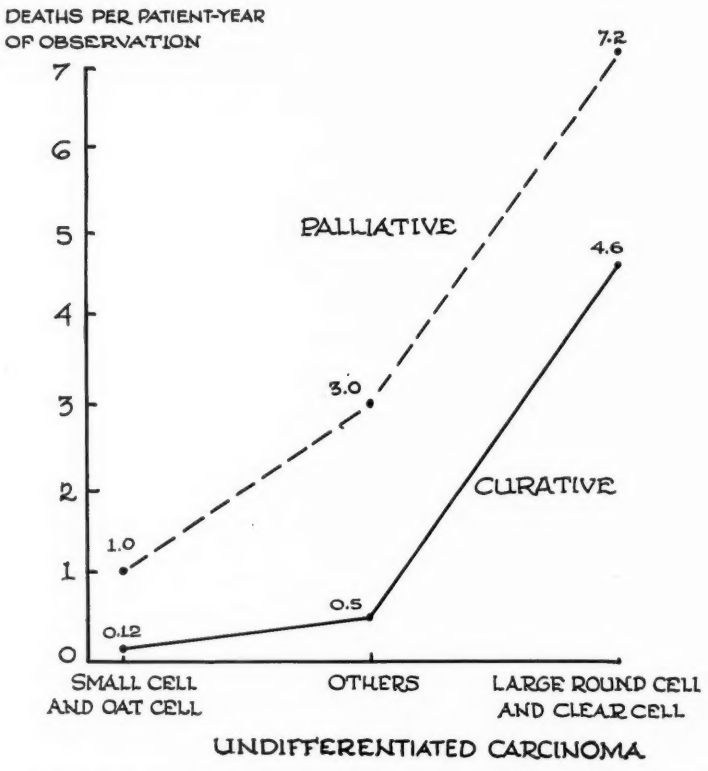

Fig. 22.—Fatality fate according to histologic type among the 53 palliative and 19 curative resections of undifferentiated carcinoma of the lung.

4. In this series, 70 per cent of the resections were considered palliative rather than curative since the lesions had extended beyond the confines of the lung. Palliative resections are considered justifiable because the risk is not excessive, the patient's remaining span of life is more comfortable, and the average survival period is increased.

5. In all but one of the 195 cases, the original slides were reviewed, classified, and graded without knowledge of the original diagnosis or fate of the patient. All carcinomas were placed in one of these groups: epidermoid (squamous cell) carcinoma, adenocarcinoma, or undifferentiated carcinoma. The epidermoid and the adenocarcinomas were divided into four grades of

histologic malignancy, and the undifferentiated carcinomas were arbitrarily divided into three subgroups according to cell type. Among the 195 cases of resection, carcinoma was found in 190, neurofibrosarcoma in 1, Hodgkin's disease in 1, lymphosarcoma in 1, malignant melanoma in 1, and 1 diagnosed as carcinoma at the time of operation was excluded because the original slides were not available. Among the 190 carcinomas, 75 were epidermoid, 43 adenocarcinoma, and 72 undifferentiated.

6. The gross survival rate after resection was 20 per cent at the end of three years with little change up to five years thereafter.

7. The survival rate in curative resections was considerably better than in the palliative group, but even in the latter a 10 per cent three-year survival rate was observed. Among the cases of nonresection, however, no patient survived three years.

8. Lesions in the right lung had a higher fatality rate than in the left lung. There was no difference in the fatality rates between centrally and peripherally located lesions.

9. Epidermoid and undifferentiated carcinomas occur in a higher proportion in the older age groups than do the adenocarcinomas. The distribution of histologic type is about equal among the younger age group, but among the older age group there is a preponderance of epidermoid carcinomas. With increasing age at the time of operation there is a progressive increase in fatality. A sharp increase in fatality occurs in patients operated upon after the age of 60 years.

10. Among the 190 patients with carcinoma of the lung there were 163 men and 27 women, a ratio of about 6 to 1. The most frequent type of tumor in men was epidermoid carcinoma whereas in women it was adenocarcinoma. The survival rate over a five-year period was consistently more favorable in women.

11. The most favorable survival rate was found in patients with epidermoid carcinoma and the poorest in those with undifferentiated carcinoma. Histologic grading of epidermoid and adenocarcinoma showed a good correlation with fatality. Among the undifferentiated carcinomas the small-cell and oat-cell tumors carried the lowest fatality and the large round- and clear-cell tumors the highest.

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CONGENITAL TRACHEOESOPHAGEAL FISTULA WITHOUT ESOPHAGEAL ATRESIA

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CONGENITAL tracheoesophageal fistula in the presence of an otherwise normal esophagus is of rare occurrence. Isolated reports of a congenital tracheoesophageal fistula as a sole entity are to be found in the literature, but most esophageal malformations consist of an esophageal atresia which is usually associated with a tracheoesophageal fistula. Largely because of its rarity, but partially because of difficulties in establishing the diagnosis of congenital tracheoesophageal fistula without esophageal atresia, the only reported instance of an attempt to correct the anomaly is the one described by Imperatori.[1] In his case, closure could be obtained through a cervical tracheotomy incision because of the high location of the fistula. In the following case an extrapleural approach was used for the closure of a congenital tracheoesophageal fistula situated within the thorax.

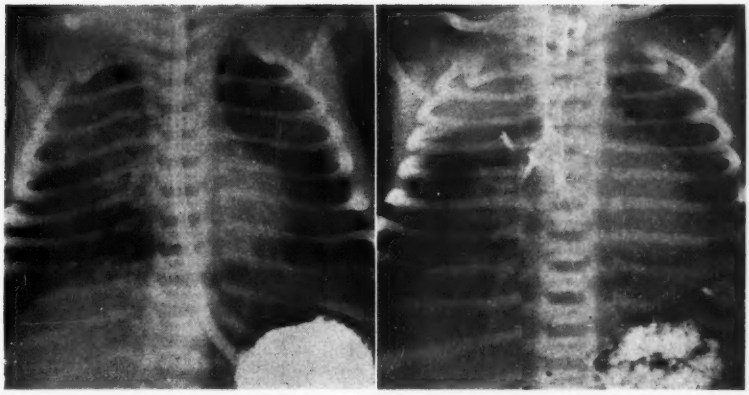
CASE REPORT

J. C. (No. 599481), male, aged 4 years, was referred to the University of Michigan Hospital on Sept. 16, 1946, by Dr. Duane Carr, Memphis, Tenn., who had established the diagnosis of congenital tracheoesophageal fistula without an esophageal atresia. The child presented symptoms of choking and cyanosis concurrent with feedings since birth. Because of the suspicion of congenital atresia of the esophagus, a catheter was introduced into the stomach on the second day after birth. Roentgenologic verification of the patency of the esophagus was obtained on the fourth day after birth, at which time barium was injected through a catheter (Fig. 1, *A*). On the twelfth day a barium suspension given orally revealed the contrast medium in the esophagus, trachea, and right bronchi (Fig. 1, *B*). It was presumed that the barium had entered the trachea by the supraglottic route and the diagnosis of tracheoesophageal fistula was not made at that time. Atelectasis of the upper lobe of the right lung was evident in the film studies on the twelfth day; the ensuing films of the chest at intervals during the next four years revealed varying degrees of bilateral pneumonitis (Fig. 2). At no time during this period were the lungs free of pneumonitis on roentgen examination. A chronic cough appeared shortly after birth; at times during the day, but more so at night, the cough was accompanied by choking and cyanosis. The symptoms occurred suddenly and with sufficient severity so that the mother had to be in constant attendance throughout the day and night; often it was necessary to invert the patient quickly in order to drain secretions from the bronchial tree.

Gavage feedings were begun on the second day after birth and were continued until the end of the first year of life when feedings were started by the oral method. The feedings often caused choking and coughing. As a result of these symptoms, vomiting frequently occurred during or shortly after meals. Solid foods were ingested more readily than liquids, and milk caused fewer symptoms than water. Because of the almost invariable choking on swallowing water, the child had an aversion to it and rarely drank it. During the period of four years, there had been seven attacks of pneumonia, and paracentesis of the m. tympani had been required on fourteen occasions because of bilateral otitis media.

From the Department of Surgery, University of Michigan Medical School.

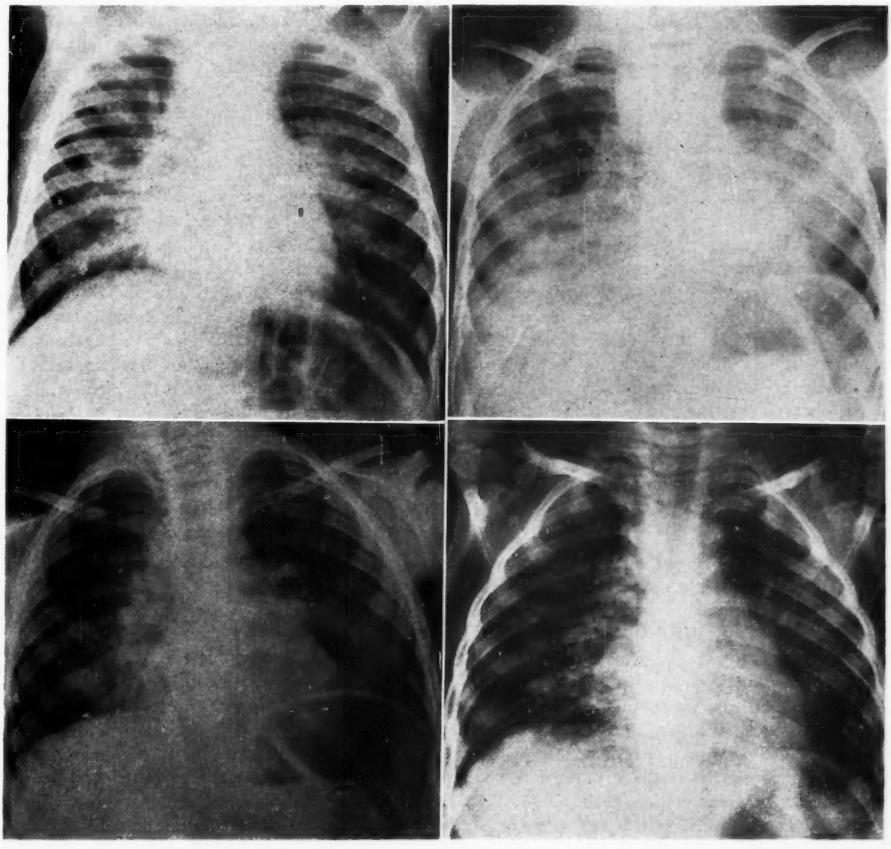
Read (by title) at the Twenty-seventh Annual Meeting of The American Association for Thoracic Surgery, St. Louis, Mo., May 28, 29, and 30, 1947.



A. *B.*

Fig. 1.—*A*, The absence of an esophageal atresia demonstrated by passage of a catheter into the stomach four days after birth. *B*, Roentgenogram following ingestion of barium twelve days after birth. The contrast medium in the trachea and bronchi was believed to have been aspirated through the larynx. (Courtesy of St. Joseph's Hospital, Memphis, Tenn.)

A. *B.*



C. *D.*

Fig. 2.—*A*, *B*, *C*, and *D*, Roentgenograms at age of 13 months, 2, 3, and 4 years, respectively, reveal the presence of varying degrees of bilateral pneumonitis. (*A*, *B*, and *C*, Courtesy of Methodist Hospital, Memphis, Tenn.)

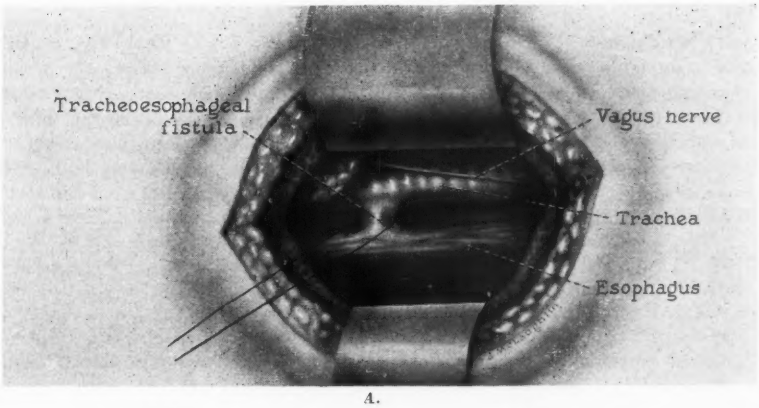
When the patient was 3¼ years of age he was seen in consultation by Dr. Carr. Since the mother mentioned at that time that the symptoms were more pronounced when the child was turned into the prone position, Dr. Carr suspected the presence of a tracheoesophageal fistula and suggested an examination of the esophagus with iodized oil and with the patient in the prone position. The oil entered the bronchial tree, outlining the trachea and both main bronchi, whereupon a diagnosis of congenital tracheoesophageal fistula was made. Operative correction of the fistula was advised but the parents did not accept this recommendation until nine months later.

On examination the child was well developed and well nourished in spite of the respiratory difficulties which had attended the feedings. The weight was 26 pounds. Respiratory wheezes were audible at a distance, and a frequent cough was productive of mucoid secretion. The nose and throat revealed evidence of an acute upper respiratory infection. The chest presented an emphysematous appearance; the breath sounds were harsh throughout and numerous râles were audible bilaterally, particularly at the bases of the lungs. The examination otherwise was not of interest. The laboratory examinations showed no deviations from normal.

Roentgenologic examination of the esophagus was made by Dr. John F. Holt on Sept. 17, 1946. Iodized oil was given orally with the patient in the *prone* position. It was observed by fluoroscopic examination that the oil entered the trachea coincident with the first swallow. The exact site of the fistula could not be determined at the fluoroscopic examination or in the subsequent films. There appeared to have been slight regression of the bilateral pneumonitis since the last preceding film studies nine months earlier. Operation was deferred for ten days after admission in order to await clearing of the acute upper respiratory infection. During this interval the child received sulfadiazine orally and penicillin intramuscularly.

Operation was done Sept. 27, 1946. Bronchoscopy, esophagoscopy, and repair of congenital tracheoesophageal fistula were carried out. After the induction of ether-oxygen anesthesia, the trachea was examined with a 4 mm. bronchoscope, but a fistula could not be seen. A 5 mm. bronchoscope was then used for examination of the esophagus. The anterior wall of the esophagus was carefully investigated, but no fistula could be found. Because the determination of the exact level of the fistula had not been possible on roentgenologic examination, and because it was believed that this information was of importance in planning the level of the operative approach, the tracheoscopy was repeated using a 5 mm. bronchoscope. The improved visibility of the trachea with this instrument resulted in the prompt recognition of a large fistulous opening on the right posterolateral wall of the trachea at a level approximately 1.5 to 2 cm. above the bifurcation. At a distance of several milimeters from the wall of the trachea, the lumen of the fistula narrowed to a diameter of 3 to 4 mm. Air and watery fluid bubbled through the fistula coincident with the expiratory phase of respiration, indicating a communication with the esophagus. The bronchoscope was withdrawn, an intratracheal tube was inserted, and the patient was then placed in the left lateral position.

The site of the incision was infiltrated with ¼ of 1 per cent solution of Metycaine hydrochloride. A right parascapular incision was made from the level of the second to the sixth ribs. The fourth and fifth ribs were resected posteriorly, the fourth intercostal muscle bundle was divided, and the parietal pleura was separated from the thoracic wall until the posterior mediastinum was entered. The esophagus and trachea were exposed and the vagus nerve was infiltrated with the local anesthetic solution. Between the lower portion of the trachea and the esophagus, a gap of about 3 mm. was present. At this level, a fistula was not evident between the two structures. The dissection was carried upward, and the tracheoesophageal fistula was encountered at a distance of about 2 to 2.5 cm. above the bifurcation. The length of the fistula was about 4 mm.; its breadth could not be ascertained until the areolar and fatty tissue adjacent to its superior edge had been reflected upward, whereupon it could be seen that the breadth was about 6 mm. A silk thread was passed around the fistula to provide traction (Fig. 3, *A*). It was apparent at this time that the exposure was insufficient to allow satisfactory closure of the fistula. The posterior portion of the third



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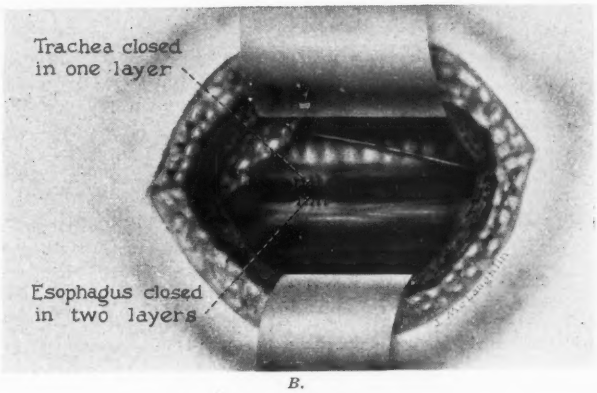

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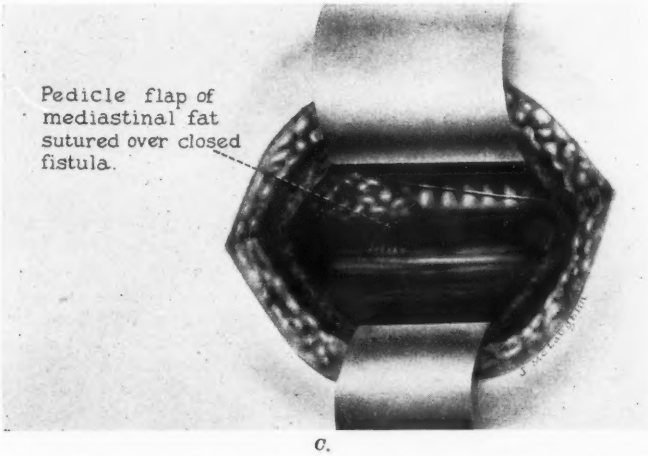


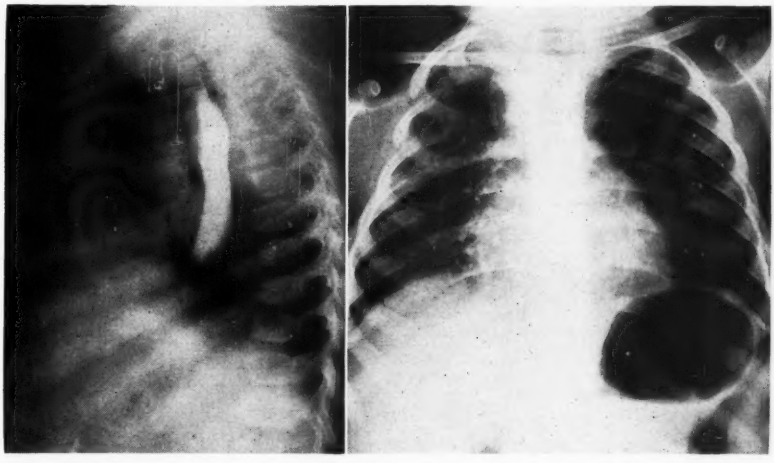
C.

Fig. 3.—*A*, The appearance of the tracheoesophageal fistula as seen on extrapleural exposure of the anomaly. *B*, The fistula has been transected close to the esophageal wall. The tracheal opening has been closed with one layer of interrupted sutures and the esophageal opening with two layers of sutures. *C*, A pedicle flap of mediastinal fat and areolar tissue has been sutured over the closed tracheal fistula in order to reinforce the suture line and to interpose a barrier of tissue between the closed fistulas.

rib was therefore resected and the third intercostal muscle bundle posteriorly was divided between ligatures. The fistula was partially transected from below upward for a distance corresponding to about one-third of its breadth. As it was anticipated that closure of the tracheal end of the fistula would be more difficult than closure of the esophageal end, the incision was placed in close proximity to the esophageal wall in order to leave a cuff of the fistulous wall attached to the trachea. This plan was thought to be necessary in view of the relatively large diameter of the fistula as it entered the trachea, as had been observed during the tracheoscopic examination. On incision of the fistula, it appeared that the horizontal diameter of the tracheal opening was slightly greater than the vertical diameter. In spite of the cuff of the fistulous wall which had been preserved, some difficulty was experienced in the placement of sutures along the inferior margin of the tracheal end of the fistula. This condition resulted from the fact that a portion of the fistulous opening extended lateral to the membranous portion of the tracheal wall and arose from the rigid cartilaginous wall. The fistula was then completely transected, and the tracheal opening was closed with approximately six sutures of interrupted 00000 silk on atraumatic needles. The opening into the esophagus was readily closed with sutures of a similar material; the first layer of sutures of an end-on mattress type was placed in the mucosa and a satisfactory inversion of its edges was obtained. The longitudinal fibers of the muscularis, which had been displaced laterally by the presence of the fistula, were then approximated to provide for a second layer of the closure (Fig. 3, *B*). In order to strengthen the closure of the tracheal end of the fistula and to interpose a layer of tissue between the tracheal and esophageal suture lines, a pedicle flap of the mediastinal fat and areolar tissue which had been displaced upward on exposing the fistula was sutured downward to cover the tracheal suture line (Fig. 3, *C*). The wound was irrigated with physiologic saline solution and 200,000 units of penicillin were instilled. As a small opening had been made in the mediastinal pleura immediately behind the junction of the azygos vein with the superior vena cava, the inner end of a narrow gauze drain was placed over the site of the pleural opening. The outer end was brought out through the central portion of the incision. Inflation of the lung was maintained with positive pressure during the closure. The anterior ends of the divided intercostal muscles were sutured to the undersurface of the sacrospinalis muscle, the extracostal muscles were approximated in layers, and the subcutaneous tissues and skin were closed in separate layers. During the operation the patient received 200 c.c. of blood by infusion into the saphenous vein at the level of the left ankle. Examination of the chest at the conclusion of the operation revealed breath sounds of normal intensity throughout the right lung and no evidence of pneumothorax.

The postoperative course was satisfactory and without complications. A Levine tube was placed into the stomach to prevent gastric distention and vomiting, but constant suction was not necessary. Fluids were administered by subcutaneous infusion and rectal instillation for the first two days after operation and were then given by gavage from the third to fifth days, inclusive. Fluoroscopic examination with the aid of iodized oil and with the patient in the prone position on the fifth day revealed no evidence of leakage of the esophageal suture line, and oral feedings were begun on the following day. Penicillin by intramuscular injection was continued throughout the postoperative course, and sulfadiazine was begun on the third day coincident with gavage feedings. The gauze drain in the extrapleural wound was shortened daily and was completely removed on the sixth postoperative day. Film studies of the chest on the first and second days revealed increased density in the extrapleural wound space, but no effusion was present in the right pleural cavity. An examination of the esophagus with barium on the twelfth day showed no abnormality (Fig. 4, *A*). At this time the pneumonitis in the right hilar region had decreased, but the pneumonitis in the left lung remained unchanged. The patient was discharged from the hospital on the thirteenth day; the wound was healed, feedings were being taken without symptoms, and the chronic cough which had been present before operation had almost completely disappeared. There was a complete absence of the râles which had been present throughout both lung fields on admission.

A roentgenogram of the chest five weeks after operation showed considerable clearing of the pneumonitis bilaterally (Fig. 4, *B*). The patient had no symptoms after his return home. The complete disappearance of the cough was particularly gratifying, inasmuch as it had been suspected that an irreversible bronchiectasis might have developed as a result of the prolonged pneumonitis. Reports from the parents at intervals during the eight months which have elapsed since operation state that the patient continues to have no symptoms.



A. *B.*

Fig. 4.—*A*, Roentgenogram twelve days after closure of tracheoesophageal fistula. *B*, Considerable clearing of the bilateral pneumonitis is evident five weeks after operation. (Courtesy of Dr. J. Cash King, Memphis, Tenn.)

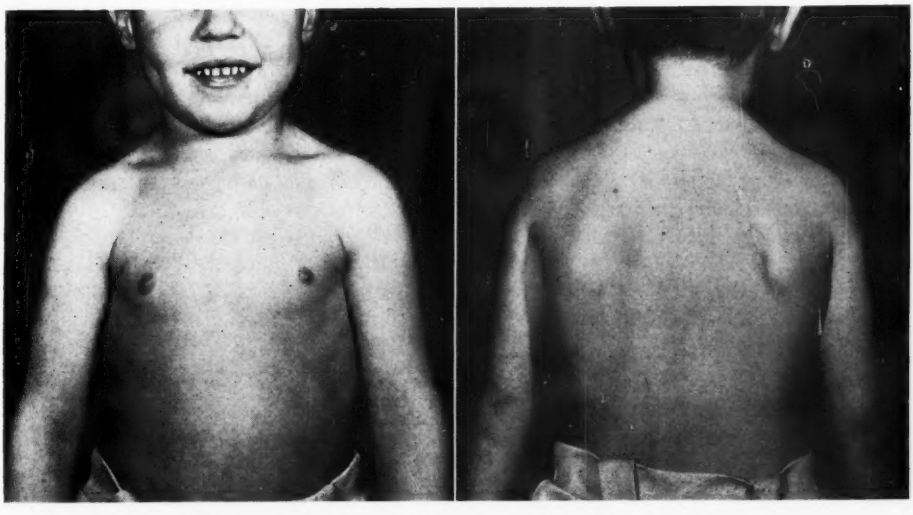


Fig. 5.—Photograph of patient five months after operation.

DISCUSSION

The large majority of congenital esophageal malformations consist of an atresia which is usually associated with the fistulous communication between the trachea and lower esophageal segment. Less often an atresia of the esophagus with varying degrees of agenesis of the lower esophageal segment and without a tracheoesophageal fistula is seen. During the past twelve years, sixty-three infants with atresia of the esophagus have been observed at the University of Michigan Hospital. In contradistinction to the frequency of esophageal atresia, only two patients have been seen with congenital tracheoesophageal fistula in the presence of an otherwise normal esophagus. One of these patients has been referred to in previous publications[2, 3] and has been illustrated by Patten.[4] The other case is the subject of the present report. The incidence of tracheoesophageal fistula as a single entity, as contrasted with the incidence of esophageal atresia, in the cases seen personally to date is approximately 3.0 per cent.

Evidence of the rarity of congenital tracheoesophageal fistula without other abnormalities of the esophagus is substantiated by a review of the literature, although discrepancies are to be found in the earlier reports of this anomaly. Cautley[5] in 1917 referred to six instances of tracheoesophageal fistula without other esophageal abnormalities in the cases of congenital anomalies of the esophagus reported previous to that date. Two years later, Plass[6] was able to find only one case of congenital tracheoesophageal fistula in 136 reported cases of malformations of the esophagus. Cautley and Plass agree in their citation of the case reported by Lamb[7] in 1873 that it was a case of congenital tracheoesophageal fistula. It has likewise been possible for me to verify the authenticity of this case, which appears to have been the first case of congenital tracheoesophageal fistula unassociated with other abnormalities of the trachea or esophagus to be reported in North America. The case of Eppinger's,[8] mentioned by Cautley as a congenital tracheoesophageal fistula and not included in the cases collected by Plass, has been found to be a congenital tracheoesophageal fistula situated just above the bifurcation of the trachea. The esophagus showed no other abnormality. Another case included by Cautley as a case of congenital tracheoesophageal fistula, but mentioned by Plass as a case of congenital atresia of the esophagus with tracheoesophageal fistula, is one described by Vrolik.[9] Eppinger described this case as having a narrow but passable area in the upper esophagus and a tracheoesophageal fistula above the stenosis. The case described independently by Pinard[10] and by Tarnier[11] was mentioned by Cautley as a case of congenital tracheoesophageal fistula and by Plass as a case of congenital atresia of the esophagus. On consulting the original references this case has been found to have had a high tracheoesophageal fistula situated immediately below the glottis. The esophagus was otherwise normal. Richter's[12] case, included as a case of congenital tracheoesophageal fistula by Cautley, is mentioned by Plass as being a peculiar abnormality whereby the trachea and esophagus formed a single sac. I have not been able to locate the original descriptions of this case and the one of Von der Water's[13] which is mentioned by Cautley as a tracheoesophageal fistula and is not included in the cases collected by Plass.

More recent reports of congenital tracheoesophageal fistula are described by Imperatori[1] and by Gengenbach and Dobos[14] (two verified cases and one presumptive case), and additional cases are mentioned by Leven and Lannin[15] (two cases), Shaw[16] (one case), and Barkley[17] (one case). The patient operated on by Imperatori was the only one for whom operation has been undertaken to correct the anomaly. In view of the fact that approximately 500 cases of congenital atresia of the esophagus have been reported to date, it is apparent that the incidence of congenital tracheoesophageal fistula as a sole entity is rare.

The symptoms of congenital tracheoesophageal fistula unassociated with other anomalies of the esophagus consist of choking and coughing coincident with the ingestion of feedings and particularly with the ingestion of liquids. In the case described in this article, the symptoms occurred especially during the ingestion of water. This finding was also observed by Clerf, Cooley, and O'Keefe[18] in an adult with an acquired esophagobronchial fistula. These symptoms occurring in relation to feedings appear to vary inversely with the viscosity of the feedings. Similar symptoms, often associated with cyanosis and at times respiratory arrest, may occur after a variable interval following feedings. Such symptoms are usually the result of regurgitation of feedings and their sudden entrance into the tracheobronchial tree in large amounts. The delayed symptoms following meals are more likely to occur if the infant is in a prone position, inasmuch as this position causes the fistula to be dependent in its relation to the esophagus and thereby facilitates the entrance of esophageal contents into the trachea. A chronic wet cough and fever occur as a result of a pneumonitis which develops sooner or later because of the repeated aspiration of foreign material into the tracheobronchial tree.

In newborn infants the symptoms of congenital tracheoesophageal fistula are not unlike those resulting from a faulty neuromuscular coordination of the act of deglutition. In both these conditions the infant can swallow a normal amount of the feeding providing the symptoms are not too severe. It is important, therefore, that these two conditions be differentiated from each other by appropriate examinations to determine the presence or absence of a fistula: If the symptoms of congenital tracheoesophageal fistula are severe, they may simulate the more common anomaly of congenital atresia of the esophagus with tracheoesophageal fistula. The diagnosis of esophageal atresia can be readily ascertained by the detection of the atresia on roentgen examination of the esophagus with the aid of a catheter or iodized oil, or the absence of an atresia can be determined by the ability to pass a catheter into the stomach.

Leven and Lannin have remarked that the symptoms of congenital tracheoesophageal fistula may be inconstant because of the small size of the fistulous track and that the diagnosis may be difficult to establish before autopsy. Unlike esophageal atresia with tracheoesophageal fistula, the symptoms of congenital tracheoesophageal fistula may not be of sufficient severity to prompt an early diagnosis. As the infants may be able to maintain their nutrition, the condition may continue to remain unrecognized and untreated, masquerading under the impression of an acute recurring pneumonitis or a faulty mechanism of degluti-

tion. In the patients with congenital tracheoesophageal fistula who have survived more than a few days, a recurring pneumonitis has been the reason for the frequent periods of hospitalization.

The length of life has varied considerably in the patients for whom operation has not been done. The shortest survival was the case described by Pinard and Tarnier in which death occurred thirty-six hours after birth and followed the operative repair of an imperforate anus. One of the patients described by Gengenbach and Dobos lived four days. Barkley's and Shaw's patients died at the age of 5 and 13 days, respectively. Lamb's patient was 7 weeks of age at the time of death. The first patient seen in the University of Michigan Hospital died at the age of 3 months. Another of the patients described by Gengenbach and Dobos was 2 years old at the time of their report and a third patient of theirs, in whom the diagnosis was suspected but not definitely established, was 8 months of age when reported. Eppinger's case was that of a "young man" whose age at the time of death was not specified. The two patients for whom operation was done were 4 years of age (the case reported in this article) and 6½ years of age (Imperatori's case) at the time of their respective operations.

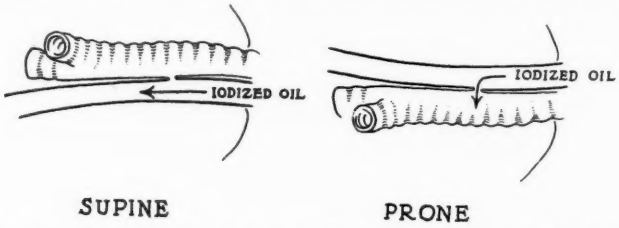


Fig. 6.—The roentgen diagnosis of congenital tracheoesophageal fistula may not be apparent if the patient is in the supine position. The prone position causes the fistula to be dependent in its relation to the esophagus, thereby allowing the iodized oil to enter the trachea by gravitation.

The diagnosis of tracheoesophageal fistula without esophageal atresia has been difficult to establish in the cases reported to date. This has been true even when the anomaly has been suspected. The diagnosis should be readily evident, however, if certain precautions are observed in the roentgenologic examination or if endoscopic examination of the trachea or esophagus or both is performed. In the first patient seen in the University of Michigan Hospital, fluoroscopic and roentgenographic examinations of the esophagus were carried out on two successive days because of the suspicion of tracheoesophageal fistula and the inability to confirm the diagnosis at the first examination. These examinations were made with the patient in the supine position in which the fistula was nondependent in its relation to the trachea. On both examinations there was no evidence of entrance of the iodized oil into the trachea. In view of these negative examinations, endoscopic examination was not done, and the diagnosis was not made until the post-mortem examination, when it was obvious that the fistula should have been detected had the fluoroscopic examinations been made with the infant in a prone position. The importance of roentgenologic examination with the patient in the prone position in cases of suspected tracheoesophageal

fistula, either of the uncomplicated congenital type or of the acquired type secondary to a repair of a congenital atresia of the esophagus with tracheoesophageal fistula, has been stressed by Holt, Haight, and Hodges.

A suggestive but not diagnostic roentgenologic sign in infants with tracheoesophageal fistula but without esophageal atresia is the presence of an unusually large amount of air in the stomach or intestines. This finding was noted by Lamb, who mentioned that the most prominent symptom in his case was "flatulence which was abundant and very painful." Although the presence of a large amount of air in the stomach is entirely physiologic in newborn infants, the amount of air observed in our first patient with tracheoesophageal fistula alone was greater than in any of the patients with esophageal atresia associated with a tracheoesophageal fistula.[3] In several patients who have undergone repair of an esophageal atresia with a tracheoesophageal fistula, the appearance of gastric dilatation has been observed coincident with the development of a recurrent tracheoesophageal fistula. The gastric dilatation becomes evident when the infant cries or strains, appearing to be due to an increase of intratracheal pressure in the presence of a closed glottis and the expulsion of air into the esophagus and stomach. It must be assumed that there is simultaneous closure of the cricopharyngeus muscle at the entrance of the esophagus during crying or straining, inasmuch as the air is forced downward into the stomach, rather than being expelled through the mouth.

The diagnosis of congenital tracheoesophageal fistula has been made by endoscopic examination in two of the previously reported cases. In the patient described by Imperatori the diagnosis was established first by C. L. Jackson during bronchoscopic examination when the patient was 6 years, 5 months of age. Tracheoscopic and esophagoscopic examinations have been employed by Holinger to establish the diagnosis in one of the cases reported by Gengenbach and Dobos. In cases with a suspected congenital tracheoesophageal fistula, more frequent use of this diagnostic method should be made. Should there be difficulty in identifying the fistula, the bronchoscopic examination should be repeated after a preliminary swallow of a colored solution, such as methylene blue, as suggested by Abbott.[19]

A tracheoesophageal fistula of congenital origin should be corrected by appropriate measures as soon as the diagnosis has been made. The pneumonitis that is usually present is an urgent indication for, rather than a contraindication to, surgical treatment. This conclusion has been arrived at as a result of personal instances of recurrent tracheoesophageal fistula in the presence of a patent esophagus following the repair of a congenital esophageal atresia with an associated tracheoesophageal fistula. In one such patient who had lived to the age of 25 months, operation had been deferred pending an improvement of the pneumonitis. This did not occur and the patient died of the pulmonary complication. In view of this experience, the surgical closure of the fistula in the case reported in this article was done in spite of the pneumonitis, inasmuch as it was believed that the only satisfactory method to cure the pneumonitis was to eliminate its cause.

The successful closure of acquired fistulas between the esophagus and the trachea or bronchi in adults has been reported by several authors. Judd, Clagett, Havens, and Moersch[20] used a cervical incision for the closure of a high tracheoesophageal fistula resulting from a bullet wound. Adams and Mabrey[21] employed a transthoracic approach for a low tracheoesophageal fistula following a nonpenetrating crushing injury. Clagett, Payne, and Moersch[22] have reported the transthoracic closure of an acquired esophagobronchial fistula in an adult with calcified lymph nodes in the vicinity of the fistula. Clerf[23] and Clerf, Cooley, and O'Keefe[18] have successfully used beads of silver nitrate or sodium hydroxide crystals fused to a carrier and applied through an esophagoscope for the closure of esophagobronchial fistulas in adults. In the case reported in this article, operative correction was elected in preference to the use of a caustic because of the need for a prompt and definitive result in view of the existing pneumonitis and because of uncertainties regarding the satisfactory application of a caustic through a small esophagoscope. Although operative correction had been planned at the time of the endoscopic examinations, it was believed, furthermore, that the size of the fistula as observed during tracheoscopy was so relatively large in comparison with the size of the trachea, that chemical closure of the fistula would have presented less chance of success than that obtained by operation.

The surgical approach to intrathoracic tracheoesophageal fistulas is readily possible by either an intrapleural or an extrapleural exposure of the anomaly. The extrapleural route was favored in the case presented in this article because of the possibility of postoperative leakage of the suture lines and the lessened chance of contamination of the pleural space should leakage occur. Simple ligation of the fistula in continuity was deemed inadvisable in view of the ease with which a ligature will cut through a fistula, re-establishing its patency. This occurrence has been observed by Ladd[24] and by Leven and Lannin in patients with congenital esophageal atresia and tracheoesophageal fistula for whom ligation of the fistula was done. Division of the fistula between ligatures was considered inadvisable for the same reason. The preferable method was believed to be a transection of the fistula and closure of the openings with sutures.

The important technical factors in the transection and closure of a tracheoesophageal fistula are the use of suture material of fine size, the preservation of a cuff of ample length on the tracheal side of the fistula, and the use of a pedicle flap of tissue which supports the tracheal closure and serves as a barrier between the closed fistulas. The suture material of preference is fine arterial silk on atraumatic needles. Because of the rigidity of the tracheal wall, closure of the opening into the trachea is more difficult than closure of the esophageal opening. An intentionally long fistulous cuff should be left attached to the tracheal side of the fistula, if possible, to prevent tension upon the suture line. This was accomplished in the case reported in this article by dividing the fistula close to its esophageal end. Even so, the length of the cuff permitted the insertion of only a single layer of sutures. In this case, it would have been preferable to have

divided the fistula flush with the esophagus so that the tracheal cuff would be of maximal length. Should the tracheal and the esophageal walls be in intimate contact above and below the fistula, as was determined at autopsy in the first patient seen in this hospital and for whom operation was not undertaken, a cuff could be fashioned by a circular incision through the esophageal wall adjacent to the fistula, thus leaving this portion of the esophageal wall attached to the trachea. The closure of this cuff should facilitate the closure of the tracheal opening without tension; the ample size of the remaining esophageal lumen and its ready distensibility should not cause sufficient narrowing to produce symptoms. The esophageal closure is readily accomplished because of the laxity of the esophageal wall, the ease with which the mucosa can be inverted, and the availability of the muscularis as a second layer of closure. A pedicle flap of mediastinal fat and areolar tissue is advantageous in buttressing the tracheal closure. Without the use of a pedicle flap, the closed fistulas remain in proximity to each other and should leakage of one occur, there is considerable likelihood of opening of the other with a resulting recurrence of the tracheoesophageal fistula. Drainage of the extrapleural wound is considered advisable in view of potential contamination, inasmuch as an aseptic method of closure is not attempted.

SUMMARY

Congenital tracheoesophageal fistula without an associated atresia of the esophagus is of rare occurrence, having been observed in approximately 3 per cent of a series of sixty-five cases of anomalies of the esophagus observed at the University of Michigan Hospital. The diagnosis may not be apparent on roentgen examination with a contrast medium, unless the examination is carried out with the patient in a prone position. Tracheoscopy, esophagoscopy, or both should be used in cases in which the anomaly is suspected. Because of its rarity and past difficulties in establishing the diagnosis, operative correction has been employed in only one previously reported patient. The case of a 4-year-old child for whom the fistula was transected and the tracheal and esophageal openings were closed by suture is presented.

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SPONTANEOUS RUPTURE OF THE ESOPHAGUS

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PERFORATION of the esophagus for any reason or at any level is always a grave lesion and one which potentially, at least, is very dangerous to life. So universally pessimistic has the outlook been that even as late as 1939 Wagner[72] was quoted for the following statement: "This tragic fate (death from esophageal perforation) can almost never be averted by surgical intervention, even if the surgery is carried out by the most skillful surgeon." Little difficulty is ordinarily encountered in making the diagnosis of perforation of the esophagus from foreign body or following instrumentation because of the antecedent history, while modern experience has amply demonstrated that prompt surgical intervention[55] plus chemotherapy and proper handling may save the lives of the majority of these unfortunates.

Spontaneous rupture or perforation of the esophagus is more dramatic in its onset and considerably more tragic in its outcome than the smaller perforations, because of the size of the opening, the widespread contamination, the shock, pulmonary collapse, emphysema, and tension pneumothorax which follow it. It is all the more lethal because the origin of the difficulty is almost never suspected as there is usually nothing to suggest an esophageal lesion, and the patient is frequently in such desperate condition as to be unable to tolerate any surgical procedure even if recommended.

The clinical picture of spontaneous rupture of the esophagus is one which is not generally recognized. The majority of the cases thus far reported in the literature, now more than fifty, have not been diagnosed before death, and in many instances not even suspected before the post-mortem examination, yet the clinical picture is so characteristic that in at least two instances (Wangensteen[74]) and our Case 5 (T. J. K.) the correct diagnosis has been made over the telephone. Since the first classical description of the condition, by Boerhaave[8] published in 1724, the case of Baron de Wassenaer, Grand Admiral of Holland, there have been a number of isolated reports of the occurrence of this condition. Excellent collective reviews have been made by Fitz[22] in 1877, McKenzie[44] in 1884, Walker[73] in 1914, Smead[66] in 1931, Girard[24] in 1934, Ridgway and Duncan[61] in 1937, Terracol and associates[69] in 1938, and in the excellent articles by Barrett[4] in March of 1946, and Eliason and Welty[21] in August, 1946, to which the reader is referred. Since that time additional cases have been reported by Foggitt,[23] Cummings,[16] Culver and Clark,[15] and an additional case by Barrett,[5] to which we can add five cases which have come under our observation.

The clinical picture in spontaneous rupture of the esophagus may vary slightly, but seems to follow rather characteristic lines. An apparently healthy

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individual (males 84 per cent), frequently robust and a notoriously heavy eater, often addicted to the use of alcoholics, suddenly begins to vomit, spontaneous or induced, sometime after eating a large meal, and suddenly develops excruciating pain, usually described as epigastric, extending through the left side of the chest to the back, followed by collapse and shock, and soon by dyspnea and cyanosis. The vomiting may continue but usually ceases and the patient develops marked thirst. Frequently emphysema (60 per cent) of the tissues at the base of the neck appears early though it may be somewhat delayed. The pain is intense, persistent, and does not respond to large doses of morphine. Occasionally there is no history of a preceding large meal, the vomiting occurring because of a neurogenic lesion, postoperative nausea, or some other condition. Sometimes there is no true vomiting, merely severe and repeated retching or rarely still there is no vomiting or retching whatsoever, perforation occurring apparently as a primary condition[61] as in Case 5. The material ejected during vomiting or retching may be food or dark brown "coffee-ground" material, occasionally associated with a small amount of blood and more rarely with free bleeding. A few of the patients present a history of previous ulcer of the stomach or duodenum[26] as Cases 1, 3, and 4; a small number have a history of mild dysphagia and substernal distress suggestive of esophagitis or esophageal ulcer, while four cases, 50, 66, 71, 77, have presented a previous history of esophageal stricture, with impaction of a bolus of food at the stricture site, the perforation occurring during a strenuous attempt to force the food past the narrowed area.

When first seen the patient is usually weak and pale, though he may be cyanotic. He is usually suffering severely, is bathed in cold perspiration, and frequently is unable to lie down. Respiration is rapid and labored, the pulse fast and weak, the temperature subnormal, and the blood pressure low. Emphysema may be present at the base of the neck, but may not develop until sometime afterward. There is frequently rigidity of the upper abdomen but not of the lower, but no tenderness or rebound tenderness.

Physical examination of the chest may be reported as negative, but frequently the findings of a pneumothorax with fluid on one or both sides, more frequently the left, may be elicited, increasing as the time following perforation increases. Almost invariably present, but frequently overlooked because only the anterior part of the chest is examined, or because the examination is not carried out in the erect position, because of the patient's serious illness, is a fluid level which may be percussed out or the succussion splash demonstrated by shaking the patient. Displacement of the heart and mediastinal structures, distant heart tones, and loss of substernal dullness as evidence of tension pneumothorax are frequently overlooked or unrecognized. The deviations of the trachea normally evident to palpation in the suprasternal notch may be obscured by the subcutaneous emphysema.

The differential diagnosis must include perforated peptic ulcer of stomach or duodenum, acute pancreatitis, mesenteric thrombosis, dissecting aneurysm of the aorta, coronary thrombosis, and perhaps pulmonary embolism and spontaneous pneumothorax from other sources. The diagnosis is most frequently

missed because the physician is unfamiliar with the condition and gives no thought to the possibilities of such a catastrophe. Meyer[50] in 1858 is credited with having made the first clinical diagnosis of the condition. Since that time apparently less than 25 per cent of the cases have been correctly diagnosed before death[4, 5, 7, 14, 16, 21, 22, 50, 73, 74] and three of ours, Cases 3, 4, and 5.

The most frequent misdiagnosis encountered in the reported cases has been that of ruptured peptic ulcer which has been made many times and at least thirteen exploratory laparotomies carried out,[2, 4, 22, 35, 45, 57, 59, 66, 73, 76, 78] including our Case 1. Mediastinal emphysema presenting in the cervical region has been present in more than 60 per cent of the reported cases and cannot be produced by any of the other conditions listed in the differential diagnosis except possibly primary spontaneous pneumothorax, and occurs but rarely in that. The presence of hydropneumothorax, either unilateral, usually the left, or bilateral,[4, 8, 16, 19, 28, 42, 45, 50, 62] has occurred in all but seven of the reported cases,[1, 21-23, 43, 63] and cannot be produced by any of the conditions mentioned in the differential diagnosis except primary spontaneous pneumothorax, and should give a clue as to the exact condition. Pain, collapse, vomiting, hydropneumothorax, and emphysema should permit a diagnosis on historical and physical grounds alone if the clinician is cognizant of the occurrence of such a condition.

Several laboratory aids may establish or confirm the diagnosis and of these the x-ray examination is of greatest value. The earliest finding on x-ray films, probably present in all cases from the onset, if recognized, is the presence of emphysema in the mediastinum, localized at first, later more extensive, involving the whole mediastinum, and eventually fanning out as subcutaneous emphysema in the cervical region. The second important finding is the demonstration of a fluid level in the mediastinum; the third, the finding of a pneumothorax on one or both sides, most frequently on the left; and fourth, the demonstration of a fluid level within this pneumothorax pocket. A demonstration of the second and fourth of these findings, of course, requires that the films be made with the patient in proper position, usually erect so that the fluid level may be demonstrated. A fifth x-ray finding of negative diagnostic importance is, of course, the absence of subphrenic air or gas indicative of perforation of a subphrenic hollow viscus. The absence of this finding, of course, does not rule out subdiaphragmatic perforation as it is demonstrable in only 70 to 80 per cent of proved gastric and duodenal perforation. If further evidence of perforation of the esophagus is required it may be obtained by demonstration of orally administered barium or iodized oil, in the mediastinum or in the adjacent pleural cavity. The iodized oil is easier to remove from the surgical field subsequently but the risk of iodism must be remembered.

The aspiration and examination of fluid from the pleural cavity may likewise confirm the diagnosis,[2, 26, 61, 67] as in Cases 3, 4, and 5. A fluid of sourish odor resembling vomitus offers conclusive proof. At times the odor of beer or other alcoholic beverage in the aspirated fluid may confirm its origin, as may the demonstration of food particles or acid, either free or combined. These are diagnostic only if one can be certain that the needle has not been introduced into

a dilated, herniated stomach or that it has not been introduced below the diaphragm. If additional proof is necessary, the prompt appearance of some colored solution such as grape juice,[26] as in Case 4, from the pleural cavity shortly after its ingestion will establish direct communication between the esophagus and pleural cavity. Failure to demonstrate abnormal findings upon peritoneal puncture is of negative diagnostic importance, if necessary. The diagnosis then of perforation of the esophagus may be made from clinical history and physical examination and may easily be substantiated by suitable x-ray examination or confirmed in the laboratory by aspiration of characteristic material from the pleural cavity. The diagnosis should be made much more frequently for it is only by early and accurate diagnosis and prompt surgical treatment that any but an outside chance of recovery can be offered to the patient.

PROGNOSIS

The prognosis in spontaneous perforation of the esophagus is always extremely grave. As late as August, 1946, Eliason and Welty[21] made the following statement: "The mortality reported so far in spontaneous rupture of the esophagus is 100 per cent, excluding the two cases mentioned by Graham." These two patients apparently had already survived sufficiently long to develop an empyema which when drained was followed by recovery and healing of the esophageal fistulas. The size of these perforations was unstated, although the course followed might suggest small slow perforations rather than the extensive "blowout" type, present in so many of the reported cases. From the reported cases, thirteen patients died under twelve hours, twenty-four additional patients under twenty-four hours, eight under forty-eight hours, and only eight survived longer than forty-eight hours, two of them living nine and seventeen days, respectively. It would seem clinically that the early deaths must result from shock and tension pneumothorax, for few of them suffer from severe hemorrhage. The later deaths apparently result from mediastinitis and pleural sepsis as well as from mechanical factors.

MECHANISM OF PERFORATION

The mechanism and possible factors underlying spontaneous rupture of the esophagus have previously been discussed by several authors.[10, 24, 44, 81, 83] McKenzie[44] in 1884 demonstrated experimentally that the bursting pressure of the human esophagus in the cadaver varied from 5¾ to 11 pounds with an average of slightly over 7 pounds, and that rupture always occurred in a longitudinal direction. Zenker and von Ziemssen[83] also showed that a longitudinal pull of 8.5 kg. with variations from 6 to 11 kg. was necessary to disrupt the esophagus, and that the mucous membrane was somewhat stronger than the muscular coat. Since only two cases of esophageal rupture in a transverse direction have occurred,[8, 57] this work has less practical application than other experimental work. Duval[20] in 1921 demonstrated that the most certain factor in perforation of a viscus by air distention was the rapidity of its introduction rather than the total degree of pressure.

Burt[11] in 1931 working with isolated segments of human intestine under slow air distention found that the sections of the rectum withstood pressure of greatest degree with the sigmoid, ileum, esophagus, jejunum, transverse colon, cecum, and stomach rupturing the sequence at diminishing pressures. The outer layers of the esophagus ruptured at 3.49 pounds per square inch (18.2 cm. of mercury) and the mucosa at 4.07 pounds per square inch (21.3 cm. of mercury).

During the past year at the Minneapolis General Hospital one of the authors (A. J. H.) has carried on a series of investigations on fresh human cadavers in which the bursting pressure of the stomach and esophagus in situ was determined by means of slow air distention applied by a Baumanometer bulb through a tube tied in the upper esophagus above the aortic arch. The anterior chest plate had been removed and the abdomen opened, but otherwise all structures of the chest and abdomen were undisturbed except for clamping the stomach at the pylorus. Examinations were carried out at intervals from 1½ to 22½ hours after death. All save one of the studies were in adults of ages ranging up to 75 years. Under the technique used the stomach became greatly distended at first and then perforated along the lesser curvature at pressures ranging from 4 to 9 cm. of mercury (0.7660 to 1.7235 pounds per square inch at room temperature), with an average of 5.85 cm. of mercury (1.1184 pounds per square inch) for thirty-three observations. Following rupture of the stomach the clamp was applied across the stomach just distal to the cardia and pressure again applied until rupture of the esophagus occurred, at pressures ranging from 10 to 33 cm. of mercury at room temperature (1.915 to 6.3195 pounds per square inch), with an average 19.27 cm. of mercury (3.69 pounds per square inch) for twenty-nine observations.

All of the perforations produced in the esophagus were longitudinal in direction and occurred in the lower third. The only exception to these readings was observed in a child of 4 months examined two hours after death in which rupture of the stomach occurred at 12 cm. of mercury (2.298 pounds per square inch) and rupture of the esophagus at 67 cm. of mercury (12.83 pounds per square inch). These observations in part closely parallel the figures published by Burt[11] in 1931, and the last observation in an infant of 4 months is in line with Burt's statements that higher pressures are required to rupture the esophagus in children under the age of 12 than in adults. The time after death seemed to make no difference in the bursting pressure observed. Morton and Sullivan[53] who reported that in dogs there is nearly a 50 per cent decrease in the bursting pressure of the duodenum and ileum within one hour after death, but that subsequently there is little change within the next twenty-four hours. Whether or not their experimental work must be applied to the previously given figures had not been determined. Such bursting pressures in the human esophagus give little comfort to those who dilate the lower esophagus and cardia with a hydrostatic or pneumatic dilator, with pressures of 22 feet of water[71b] or more (48.537 cm. of mercury or 9.63 pounds per square inch) were it not for the fact that much of this pressure is dissipated against the rigid wall of the silk bag of the dilator rather than in stretching the esophagus or stomach beyond a certain predetermined diameter.

Can pressures equal to those necessary to rupture the esophagus in the laboratory be generated in the living human being? We have been unable to find records of vomiting pressures in any of the available literature. Healthy young adults can easily blow pressures of 110 to 125 mm. of mercury (2.11 to 2.39 pounds per square inch) while an occasional husky individual may blow 180 to 190 mm. of mercury (3.44 to 3.63 pounds per square inch). What heights can be reached by forceful, spasmodic muscular contractions of forceful vomiting or retching are unknown, but they must exceed those produced by voluntary muscular contractions which are at least equal to the minimal pressures necessary to rupture the apparently normal esophagus at the post-mortem table.

Fig. 1.—Longitudinal rupture of lower esophagus produced at post mortem by air distention at 18 cm. of mercury pressure.

Care must be exercised in attempting to apply directly the readings obtained on the experimental animal, particularly as to bursting pressures, because it is well recognized that the esophagus in the dog is apparently much stronger than that in the human being. Pressures at least as high as 70 cm. of mercury (13.4 pounds per square inch) have been found necessary to rupture the esophagus in an 18 kilogram dog. A pressure as high as 67 cm. of mercury has previously been recorded in a 4-month-old infant. Whether these readings represent an actual increase in strength of the wall of the esophagus or are merely a manifestation of the relation of wall strength to diameter of the tube according to the formula $T = II\ D\ Pr.$[74b] has not been determined. The longitudinal bursting of the esophageal wall may be related to the ease of disruption of the longitudinal fibers or may merely be another manifestation of the well-known fact that pipes usually burst in a longitudinal direction.

Clinically, spontaneous perforation of the esophagus, excluding all patients who have had recent esophagoscopy, dilatation, bouginage, or passage of a tube,

or who have rupture of an abscess or aneurysm or perforation of a carcinoma, a peptic ulcer in ectopic gastric mucosa, or other similar gross lesions, would seem to occur under several circumstances, not all of which are mechanically the same. These may be listed as follows:

1. Vomiting spontaneous or induced, shortly after the taking of a large meal
2. Recurrent vomiting and retching, which occurs after the stomach apparently has been more or less emptied
3. Perforation associated with retention or distention without vomiting or retching
4. Vomiting in the patient with a neurogenic lesion
5. Forceful attempts at swallowing or retching to dislodge impacted food in the patient with esophageal stricture and
6. Any of these in the patient with a history suggestive of esophagitis sometime prior to the perforation.

Traumatic rupture of the lower esophagus has been reported.[2-54] Spontaneous direct rupture from mechanical forces alone can be assumed in some patients while in others the possibility of some weakening of the esophageal wall must also be considered. Whether this involves weakening of the mucous membrane by infection, erosion, partial digestion, or a combination of more than one factor plus the application of a mechanical force has not been determined. Experimental studies along this line are under way but have not progressed far enough to justify reporting at this time. Certainly the microscopic demonstration of inflammatory reaction on the surface or in the esophageal wall adjacent to esophageal perforation of twenty-four to forty-eight hours standing in the presence of the very virulent infection associated with this tragedy does not of itself prove a previously existing esophageal inflammation.

From the clinical standpoint, once an esophageal perforation is diagnosed, it makes no great difference whether the rupture has occurred from pressure of vomiting through a normal structure, or whether the perforation has resulted from erosion, ulceration, dilatation, or other instrumentation except as to the extent of damage done. The perforation itself is the critical lesion which demands prompt and thorough surgical treatment no matter what its cause. Upon such treatment lies the patient's best and perhaps only chance of recovery from this catastrophe.

TREATMENT

When confronted with Case 3 of our series, with the experience gained from the misdiagnosis in Case 1 (Case 2, though earlier in time, was added to the series later), the diagnosis was promptly made from the x-ray studies by one of us (R. W. M.) and surgical consultation was promptly requested. Surgical intervention was immediately considered but postponed because of the patient's grave condition in the hope that it might be improved sufficiently to permit thoracotomy and repair. The fallacy of this hope was later realized. One patient (Case 4) was seen very late, possibly two weeks after the onset of the condi-

tion in a very old and feeble man, quite evidently in no condition for surgical intervention. The diagnosis, however, was made and the possibilities of surgical intervention evaluated.

When another patient (Case 5) presented himself, the diagnosis was made clinically, very promptly, in fact over the telephone in conversation with the surgical resident at the Minneapolis General Hospital, and the following program previously determined upon was carried out in spite of the seriousness of the patient's condition. The plan used in this patient, without knowledge of any previous attempt to treat this condition surgically, is the one which we recommend at the moment. It consisted essentially as follows:

1. Immediate treatment for shock by transfusion of blood and plasma, and electrolytes in an attempt to improve the patient's condition for surgical intervention
2. Immediate decompression of the tension pneumothorax by needle aspiration of the pleural cavity, followed by intercostal catheter thoracostomy with continuous suction drainage to prevent its recurrence
3. Discontinuation of all fluids by mouth, satisfying the patient's intense thirst by intravenous and subcutaneous fluid administration
4. Immediate institution of intensive chemotherapy with sodium sulfadiazine and penicillin added to the intravenous fluids
5. The carrying out of further diagnostic procedures to confirm the diagnosis and
6. Open posterior thoracotomy under intratracheal anesthesia with complete removal of all food and stomach contents from the pleural cavity, a wide opening of the mediastinum, and removal of all food particles and fluid from it, followed by exposure of the ruptured area of the esophagus and closure of the perforation with nonabsorbable sutures of silk, or possibly of chrome wire as advocated by Belsey[6] or even better perhaps with tantalum wire because of its lack of reaction in infected tissues.

The perforated area of the esophagus may be covered by pleura but free drainage from the mediastinal pocket through the pleura must be established and left open. The mediastinum and pleural cavity must then be thoroughly washed out with large volumes of sterile salt solution, possibly followed by irrigation with $\frac{1}{1000}$ solution of Zephiran, which, however, must not be allowed to remain in the pleural cavity, for in dogs at least it is toxic. The local implantation of penicillin, sulfonamides, and streptomycin is justifiable. The chest is then closed tightly leaving three or four catheter drains placed with one along the esophageal perforation, another into the mediastinum, another in the gutter region posteriorly, and a fourth along the posterior costophrenic angle. Continuous suction is applied to these catheters as soon as the patient is returned to his room. The lung, of course, is expanded as the chest is closed.

While the esophagus is still open the stomach contents must be evacuated by suction and a nasal feeding tube inserted through the esophagus into the

stomach, for postoperative suction. Postoperatively, continuous suction is kept on the nasal gastric tube to insure against any distention of the stomach, which might give tension on the suture line, and nothing is permitted by mouth for several days after which feedings are then started through the nasal tube. Supportive measures of all kinds are carried on with blood transfusions, intravenous fluids, oxygen, and intensive chemotherapy. Careful watch must be kept for the appearance of fluid in the contralateral pleural cavity, and early aspiration done if it appears. With this program we are confident that some of these patients may be saved.

CASE REPORTS

CASE 1.—J. H. R., a man, aged 73 years, retired, was admitted to St. Barnabas Hospital 11:50 p. m. on Feb. 1, 1943, and gave the following history. For a number of years he had had a history of chronic duodenal ulcer, but apparently had been going along without too much trouble until the night of Feb. 1, 1943, when soon after eating a large dinner he was seized with very sudden, violent abdomial pain, followed by the development of shock and collapse. He was seen in the home by his attending physician, who made a tentative diagnosis of perforated peptic ulcer, and gave him two hypodermics of ½ gr. of morphine sulfate without relief. He was brought to the hospital as soon as the transfer could be made.

He was admitted in profound shock, with a systolic blood pressure of 80 mm. of mercury, a temperature of 97.3° F., a pulse of 105, and a respiratory rate of 35. A flat film of the abdomen taken with the patient in a somewhat erect position showed no free air under the diaphragm, and an x-ray film of the chest revealed a consolidative process in the lower portions of both lung fields, more on the left than the right, suggestive of a lobular pneumonia. This film apparently was not made in the erect position. Surgical consultation was held, and the diagnosis of probable perforated peptic ulcer was accepted and preparations made for an immediate laparotomy. An abdominal exploratory operation was carried out at 1 A.M. on Feb. 2, 1943, at which time no evidence of perforation of any hollow viscus could be found, the appendix was removed, but no abnormality noticed.

Following the surgical exploration, the shock state persisted and the pulse remained weak and irregular. By 3:10 A.M. on February 2 the blood pressure was 50/30. Oxygen and intravenous fluids were administered as well as various stimulants without effect. The respiration became more rapid, the blood pressure fell gradually, and the patient died at 12:30 P.M. on Feb. 2, 1943, twenty-six hours after admission to the hospital, and approximately thirty hours after the onset of the pain.

Post-mortem examination, Feb. 2, 1943, approximately two hours after death, revealed the following pertinent findings:

The left pleural cavity contained 1,500 c.c. of dark brownish fluid of an odor suggestive of stomach contents containing fragments of recently ingested food. There was complete atelectasis of the left lung with the exception of a small portion of the upper lobe. The right pleural cavity contained 700 c.c. of sanguineous fluid, but no adhesions. The upper esophagus was normal. In the lower esophagus just above the cardia there were numerous superficial erosions of a linear type. The largest erosion adjacent to the left border had perforated through an opening 3.8 by 2 cm. The contents of the stomach apparently had dissected the wall of the esophagus about 7 cm. before penetrating through the left pleura. The stomach was full and distended, but grossly normal. There was an ulcer 1.2 cm. in diameter on the posterior wall of the first part of the duodenum, with indurated edges, indicative of chronic ulceration. There was a small superficial ulcer .7 cm. in diameter adjacent to this.

Microscopic section of the esophagus showed local destruction of the lining squamous epithelium. The submucosa showed many polymorphonuclear leucocytes and fibrin. The wall was infiltrated with a purulent inflammatory exudate. The adventitia was dissected

free from the muscle, and this pocket showed food fragments and an acute phlegmonous exudate which extended for several centimeters above the area of perforation. The stomach adjacent to the esophagus showed a chronic gastritis with the mucosa infiltrated with lymphocytes and plasma cells.

Fig. 2. (Case 1).—Post-mortem specimen; spontaneous longitudinal perforation of lower esophagus.

CASE 2.—D. O., male, aged 16 years, was admitted to the contagious department of the Minneapolis General Hospital on Oct. 23, 1943, complaining of headache, fever, pain in the back, cough, vomiting, and abdominal pain. He gave a history of having had a "cold" for about one month after a severe sore throat. This was followed by respiratory symptoms with yellowish purulent postnasal discharge, and a slight cough producing a whitish mucoid sputum in small amounts. For about two days prior to admission, he had raised a moderate amount of blood-stained sputum. For nearly five days prior to admission, he had continuous rather severe headache in the occipital region with pain extending down the back of the neck to the dorsal spinal region, and even toward the abdomen. About the same time, he lost his appetite and began to vomit. This became so severe and continuous that for five days he was unable to retain anything taken by mouth. Upon admission he was unable to void without pain, and had considerable weakness of both legs, particularly the left.

Physical examination revealed a well-developed young adult, 5 feet, 11 inches tall, weighing 170 pounds, with a temperature of 102° F., a pulse of 90, and a blood pressure of 140/98. The pharynx was injected, and there was slight cervical adenitis. The heart was not enlarged, but there was a soft systolic murmur in the left second interspace transmitted

along the left sternal border. Chest examination was reported as negative. There was general abdominal rigidity and tenderness, but no rebound tenderness. Neurologic examination revealed some cervical and back rigidity, and a general paresis more marked in the legs with some spasm in the quadriceps and gastrocnemius muscles. The general reflexes were negative except for absent adbominal and cremasteric reflexes. Superficial pain sensation was intact, but deep pain sensation was depressed.

The hemoglobin was 92 per cent, the leucocyte count 15,450 with 84 per cent polymorphonuclear leucocytes. The urine was negative, except that it was very concentrated. The spinal fluid was clear and colorless, but contained 197 cells per cubic millimeter, of which 20 per cent were neutrophiles, and 80 per cent mononuclear cells. The sugar content was 85 mg. per cent, and the protein 37 mg. per cent.

A clinical diagnosis of acute anterior poliomyelitis was made. Chest and body packs were ordered, and oxygen was given for dyspnea as the respiratory distress increased. The clinical course was increasingly downhill, and respiratory distress increased to the point where a respirator was necessary. He gradually lapsed into coma and died at 4:15 P.M. on Oct. 25, 1943. At no time were there any clinical findings to suggest esophageal perforation, although this patient was desperately ill, irrational, and later comatose. The difficulty in breathing was considered to be on the basis of poliomyelitis. No subcutaneous emphysema was noted at any time.

Post-mortem examination 2¼ hours after death revealed a slitlike perforation in the middle third of the esophagus, opening directly into the left pleural cavity. The left pleural cavity contained 500 c.c. of gastric contents containing blood. There were no adhesions present. The left lung weighed 200 Gm., and was grossly negative. The right lung and pleural cavity were negative. In the fundus of the stomach, the mucous membrane was definitely eroded, and there were several large areas of superficial ulceration about 4 by 4 cm. in diameter covered with a gelatinous mucoid coating. Microscopically the stomach showed only a post-mortem degeneration, with no pre-mortem disease. The esophagus upon microscopic section showed epithelial ulceration, with subepithelial neutrophilic infiltration. There were definite post-mortem changes present. There was no inflammatory reaction in the external layers of the esophagus. An acute superficial esophagitis was present. Microscopic sections of the brain revealed areas of perivascular neutrophilic and mononuclear infiltration, with multiple miliary foci in the gray area. Final diagnosis was acute poliomyelitis, acute superficial esophagitis. It was the pathologist's opinion at the time of the autopsy that the perforation was a post-mortem lesion, but the evidence at hand does not fully confirm his. The post-mortem examination was conducted 2¼ hours after death. The perforation was slitlike, in the middle third of the esophagus, and the pleural cavity contained gastric contents and blood. These are not the findings of the typical post-mortem digestion perforation.

CASE 3.—W. G., a 70-year-old man, was admitted to St. Barnabas Hospital at 8:15 P.M., on Nov. 27, 1944, and presented the following history: General health apparently had been quite good until March of 1943 when he developed symptoms suggestive of duodenal ulcer, and had been placed on medical management with benefit. He was then apparently relatively symptom-free until Nov. 13, 1944, when he developed nausea, vomiting, and loss of appetite. He returned to the ulcer regimen, the symptoms subsided, and though rest was advised, he insisted upon going back to work. Two weeks later on Nov. 27, 1944, at 6 P.M. he developed a sudden severe attack of abdominal pain, mostly in the epigastric region, with difficulty in breathing, and became so ill that he was removed to the hospital by ambulance, arriving two hours after the onset of the attack.

Upon admission, temperature was 98.6°F., pulse 132, respiration 40, blood pressure 90 over 50 mm. of mercury. He was complaining of very severe epigastric pain, was definitely in shock, with profuse perspiration, and was evidently critically ill. There was definite epigastric tenderness without rigidity and dullness to percussion in the left lower part of the chest. X-ray films of the abdomen with the patient in an erect position showed no free gas under the diaphragm. X-ray films of the chest revealed emphysema of

the mediastinal tissues and the soft tissues of the neck with a left hydropneumothorax. A diagnosis of ruptured esophagus or of a herniated stomach was made by one of us (R. W. M.) from the x-ray findings.

The patient was seen in surgical consultation at 10:30 P.M. on Nov. 28, 1944, at which time he was found in shock, was markedly cyanotic and dyspneic, and had a temperature of 102° F., and a pulse unobtainable at the wrist. There was subcutaneous emphysema of the tissues of the neck, and a left tension hydropneumothorax present with displacement of the heart and mediastinum to the right. A diagnosis of perforation of a hollow viscus into the left pleural cavity was made. A left pleural aspiration was carried out, and 700 c.c. of gas, and 900 c.c. of dark thin fluid without odor was aspirated. This accorded the patient rather marked relief from respiratory distress, but the shock continued. The aspirated fluid in the laboratory was found to be gastric contents with hydrochloric acid present. The hemoglobin at this time was 110 per cent, the hematocrit reading was 53 per cent red blood cells. The leucocyte count was 11,400 with 92 per cent polymorphonuclear leucocytes. The blood Rytz test was negative. Oxygen was administered.

Supportive treatment was given with adequate intravenous fluids, transfusions, and penicillin both intramuscularly and directly into the left pleural cavity, in an attempt to improve his condition sufficiently so that surgical exploration and closure of the esophageal rent could be carried out. Sodium sulfadiazine was also administered for twenty-four hours, but this was discontinued because of inadequate urinary output, which could not be increased in spite of adequate fluid intake. A transnasal gastric suction tube was inserted and continuous siphonage applied to keep the stomach empty; oral fluids were withheld. A left intercostal catheter thoracostomy was performed, and siphonage established for continuous removal of fluid from the left pleural cavity and to favor re-expansion of the lung. Additional fluid aspirated from the left pleural cavity showed a total acid of 55 degrees, without free hydrochloric acid, and cultures of this material revealed *Staphylococcus aureus*. In spite of supportive measures, chemotherapy, adequate drainage through the left pleural cavity, and repeated transfusions, the shocklike state persisted, and the urinary output remained extremely low (below 100 c.c. in twenty-four hours), the temperature mounted, and he died at 6:45 P.M. on Nov. 30, 1944, without ever improving sufficiently to permit surgical intervention.

Post-mortem examination by one of us (A. J. H.) was carried out at 9 A.M. on Dec. 1, 1944, fourteen hours after death, and revealed the following pertinent findings:

The peritoneal membranes were free and glistening. There were 50 c.c. of dark brownish fluid present. The liver weighed 1,600 Gm. and was of a yellowish-brown mottled appearance. The heart weighed 350 Gm., the coronary arteries were patent, but there was slight sclerosis present (grade 1 on the basis of 4) and with a few atheromatous plaques. The left pleural cavity contained about 800 c.c. of dark reddish-brown fluid and the pleural surfaces were covered with a yellowish exudate. The left lung was collapsed, hemorrhagic, weighed 650 Gm., and showed edema and atelectasis. The right pleural cavity contained 1,500 c.c. of dark reddish fluid; the right lung weighed 700 Gm., and was bluish-gray in color. There was decreased crepitation and complete atelectasis of all lobes.

The esophagus showed a linear perforation 3.5 by 1.2 cm. in the lower lateral portion just above the diaphragm, extending directly to the left pleural cavity. The point of perforation was covered with a large amount of bluish-gray material. The adjacent esophageal mucosa showed numerous small linear superficial erosions. The edge of the perforation was smooth and not indurated, and showed no gross evidence of carcinoma. The stomach contained some dark brown material, yet the walls appeared to be normal. There was obstruction at the pylorus which would admit only the tip of the small finger. There was a chronic duodenal ulcer 1 cm. in diameter on the anterior wall of the first part of the duodenum adjacent to the sphincter. On the posterior wall of the duodenum opposite the anterior ulcer were two small ulcers about ½ cm. in diameter. The kidneys weighed 110 and 150 Gm., respectively. The capsules stripped with difficulty, and there were a number of deep scars on the surface. The prostate was somewhat enlarged, grade 2, with one hemorrhagic area 1 cm. in diameter, but no evidence of malignancy.

Microscopically, kidneys showed no findings to account for the oliguria. Microscopic sections of the esophagus in the region of the perforation showed an acute ulceration, with suggestion of superficial erosion of the epithelium. The borders were covered with acute fibrinous exudate. The entire esophageal wall in the region of the perforation showed the microscopic picture of an acute inflammatory exudate. There was no malignancy present, and no areas of ectopic gastric mucosa.

COMMENT

This patient's oliguria was undoubtedly related to the persistently low blood pressure. In light of our present knowledge and experience, he should have been operated upon without delay. The falacy of postponing surgical exploration until the condition could be improved is well emphasized by the final result.

CASE 4.—A. A., a 74-year-old man, was admitted to Minneapolis General Hospital on Feb. 3, 1945, complaining of severe epigastric pain, vomiting of blood, inability to swallow solid foods, and a fifty pound weight loss. He gave a history of epigastric distress coming two to three hours after meals for the past ten years, and had been studied at the Mayo Clinic some five years before, where diagnosis of chronic duodenal ulcer was made, and appropriate medical measures recommended. He was seen again at the Mayo Clinic in April, 1943, where again a chronic duodenal ulcer with a large crater and some evidence of obstruction was found. He was hospitalized and the ulcer treated medically with satisfactory improvement.

He returned again to the Clinic in July, 1944, with symptoms similar to those previously described, plus the complaint of a great deal of pain in the left lower portion of the chest, aggravated upon deep breathing and by motion of the left arm. Findings with x-ray views of the chest were negative at that time except for some elevation of the left diaphragm and some tortuosity of the aorta. An x-ray of the stomach and duodenum at that time revealed a duodenal ulcer with a small crater. Because of the pain in the lower part of the chest and some history of dysphagia an esophagoscopic examination was performed, but nothing abnormal was found. On an ulcer diet, he was relieved of the symptoms. In December, 1944, he again returned to the Mayo Clinic because of some difficulty in swallowing, but it was the impression that at that time the symptoms were functional in nature. Esophageal sounds up to No. 41 French were passed without encountering any evidence of obstruction.

On Jan. 20, 1945, two weeks before admission to the Minneapolis General Hospital, he had been vomiting considerably during the day, a dark coffee-ground material containing some fresh blood. That evening while lying in bed, he was seized with a sudden very severe epigastric pain which doubled him up. After a time the pain lessened somewhat, but persisted in a moderately severe form until admission to the hospital, where he frequently vomited coffee-ground material containing some bright blood and passed black tarry stools. He had been vomiting four or five times daily for the two weeks prior to admission, was very weak and lethargic, and complained considerably of shortness of breath. There was an indefinite history of dysphagia for solid foods for several months, a history of some dyspnea on exertion for two or three years, with occasionally swelling of the lower extremities.

Physical examination upon admission, Feb. 3, 1945, revealed a somewhat senile man, very ill, with evidence of recent extensive weight loss. The pulse was 120 and of poor quality, blood pressure was 100 over 64 mm. of mercury, and rectal temperature was 101° F. He was very pale and lethargic. There was no emphysema of the neck or chest. Examination of the heart and lungs was reported as negative. There was moderate abdominal rigidity of the upper half of the abdomen, but no pain or tenderness, and but slight rebound tenderness in the upper abdominal region. A tentative diagnosis of chronic peptic ulcer with hemorrhage was made but the possibilities of carcinoma of the stomach and perforated peptic ulcer were considered. A bedside roentgenogram of the chest, not in erect position, showed an increased density in the left lower part of the chest suggestive of fluid without a fluid level and a localized accumulation of gas in this region.

By the next day his general condition had deteriorated considerably. He was very weak and cyanotic, his blood pressure had dropped to 80 over 65 mm. of mercury, and there was increased dyspnea. A left thoracentesis was performed and 1,870 c.c. of thin brownish fluid without foul odor were removed. Examination of this fluid showed 217,000 cells per cubic millimeter, of which 93 per cent were polymorphonuclear leucocytes, and 78,000 red blood cells. Cultures of the fluid revealed a pneumococcus type IV. Sulfadiazine and penicillin were instilled into the pleural cavity following the aspiration. Abdominal paracentesis revealed only an occasional polymorphonuclear leucocyte but no bacteria present. Following the aspiration and intravenous administration of fluids and blood, the condition improved slightly and color became better under oxygen therapy. On February 5, an additional left thoracentesis was performed, and 1,000 c.c. of brownish fluid were aspirated.

The patient was seen in surgical consultation on Feb. 6, 1945, and an intercostal catheter thoracostomy was carried out to relieve the increasing left tension hydropneumothorax. Perforation of the esophagus into the left pleural cavity was recognized at that time (T. J. K.), but it was quite evident that the patient was in no condition for any major surgical therapy. The fluid removed from the chest showed a pH of 5.5 and was negative for fats, urobilin, and urobilinogen. The hemoglobin upon admission was 83 per cent, the leucocyte count was 18,300, with 81 per cent polymorphonuclear leucocytes. Electrocardiographic studies showed a sinus tachycardia, an apparent left axis deviation, and marked low voltage on all leads which was interpreted as indicative of severe myocardial damage.

The diagnosis of perforation of the esophagus into the left pleural cavity was readily confirmed by administering a small amount of grape juice by mouth, and promptly recovering the same material in the fluid removed by catheter from the left pleural cavity. The clinical course was slowly and progressively downhill from the onset. The temperature varied from 100 to 104° F. per rectum, and on Feb. 15, 1945, the patient and his family, realizing fully the gravity of the situation, decided to move him to Rochester to the Mayo Clinic where he died within two hours of arrival.

Post-mortem examination performed there* revealed a clean, slitlike perforation of the lower esophagus, directly to the left pleural cavity, with little associated mediastinitis. There was no gross evidence of previous esophageal ulceration. There was also a locally perforating duodenal ulcer with hemorrhage, and a very localized associated peritonitis. Although a diagnosis of perforation of the esophagus of the left pleural cavity was made early in this patient, at no time could surgical intervention be seriously considered because of age, senility, desperate physical condition, and the apparent duration of the process before admission. Survival was unusually long for a patient presenting this serious condition, and probably was due primarily to the fact that the perforation occurred directly to the left pleural cavity with little mediastinal infection and to a lesser degree to adequate pleural drainage. From the standpoint of the local findings alone, surgical repair would have been quite simple.

CASE 5.—W. F. C., a white man, aged 67 years, was admitted to the Minneapolis General Hospital at 4:40 P.M. on Feb. 19, 1946, in shock with marked dyspnea and extensive subcutaneous emphysema of the chest and neck. The following history was obtained: Shortly after noon of the same day while walking down the street on the way to lunch, he suddenly developed very severe epigastric pain, pain in the left side of the chest, and immediately collapsed. There was no vomiting. He was taken to his home by taxicab where his family reported that he soon developed marked difficulty in breathing and increased swelling of the neck and face. A local physician was called, who tentatively diagnosed the condition as heart block and gave an intravenous injection which relieved the pain somewhat. The pain, dyspnea, and swelling gradually increased until the patient was brought to the hospital one or two hours later.

Past history was negative except for a right parietal skull fracture in 1941. His wife reported that he had some gas and difficulty with meals for a couple of years, occurring every week or so, but that he obtained definite soda relief. An x-ray study of the gastrointestinal tract a few months before, however, had been reported as negative. There had been no

*Reported through the courtesy of Dr. H. J. Moersch.

previous food dyscrasia or dysphagia. He had been in the habit of using mineral oil and laxatives for some time. There was slight nocturia one to two times per night for five to six years. There had been an indefinite history of some pleurisy-like pain in the left side off and on for some time.

Upon admission, he was suffering from marked respiratory distress, and was rational but could talk very little because of dyspnea. The skin was warm and dry, and there was marked subcutaneous emphysema of the neck, left side of the chest, and axilla. The trachea was deviated to the right. The neck veins were not distended. Blood pressure was 80/42, the pulse 140, regular but fast, and very weak. The respiratory rate was 40 per minute. Chest examination showed the right lung clear. The left side of the chest was hyperresonant throughout, with an absence of breath sounds. The heart sounds were faint, rapid, and regular. The abdomen showed no tenderness, rigidity, or masses. The diagnosis upon admission was spontaneous pneumothorax on the left, cause undetermined. X-ray view of the chest revealed a left hydropneumothorax with subcutaneous emphysema, more marked on the left side.

The diagnosis of spontaneous perforation of the esophagus was made by one of us (T. J. K.) during telephone conversation with a surgical resident at this time, with a recommendation for surgical repair made as soon as the patient could adequately be treated for shock and prepared for surgery. A small amount of barium given by mouth appeared promptly in the left pleural cavity on x-ray films. Intravenous fluids, penicillin, sodium sulfadiazine, and blood were administered to the patient in preparation for surgery. The hemoglobin was 95 per cent, the leucocyte count 7,900 per cubic millimeter, with 98 per cent polymorphonuclear leucocytes, and only 2 per cent lymphocytes. The urine showed specific gravity of 1.033, albumin 3 plus, a faint trace of sugar, and 3 to 4 red blood cells, and 4 to 6 leucocytes per high-power field in the sediment. Fluid aspirated from the thoracic cavity was of an odor suggestive of stomach contents, showed yeast and gram-positive cocci in chains on smear, and a nonhemolytic streptococcus on culture. Electrocardiographic studies revealed a sinus tachycardia with elevation of the ST in Leads II and III suggesting myocardial damage.

Surgical exploration was carried out at 10:30 P.M., approximately 8½ hours after the onset of the trouble, under intratracheal cyclopropane anesthesia, after the insertion of a nasal suction tube into the esophagus and the stomach. The chest was opened through a posterolateral incision with resection of a wide segment of the ninth posterior rib. Upon opening the pleural cavity, more than 1 liter of fluid and food was encountered with numerous peas, beans, pickles, and other undigested food remnants easily recognizable. This represented food taken twenty hours prior to the onset of the sudden attack. All solid food material was removed from the pleural cavity, which was then thoroughly irrigated with a large amount of sterile water until all foreign material had been removed. The barium which had previously been given by mouth could be observed as a thin white coating on the pleura. This was removed as thoroughly as possible. Exploration revealed a 9 cm. rent in the left pleura overlying the esophagus, and a 4 cm. long perforation on the left posterolateral wall of the esophagus just above the cardiac end of the stomach. The mucosa of the esophagus was protruding through this rent. Direct examination revealed no evidence of chronic ulceration or neoplasm in this area. A finger inserted into the esophageal perforation immediately entered the stomach, and through this opening all contents of the stomach were removed. There was no evidence of scarring or ulceration in the stomach that could be demonstrated through this exposure.

The rent in the esophagus was then closed with interrupted sutures of 000 silk, first to the mucosa, a second layer through the overlying muscle, and a third layer through the overlying pleura, leaving, however, a small communication between the mediastinum and the left pleural cavity for drainage. The lung was expanded repeatedly during the procedure. Four intercostal catheters inserted through stab wounds were left in the pleural cavity. Five grams each of sulfanilamide and sulfathiazole were then sprinkled into the mediastinum and pleural cavity, and 100,000 units of penicillin injected. The chest wall was then closed without further drainage. The patient tolerated the surgical exploration extremely well, though the blood pressure when the anesthetic was administered was only 60/40, and at the end of surgical procedure it had come up to 100 over 60 mm. of mercury.

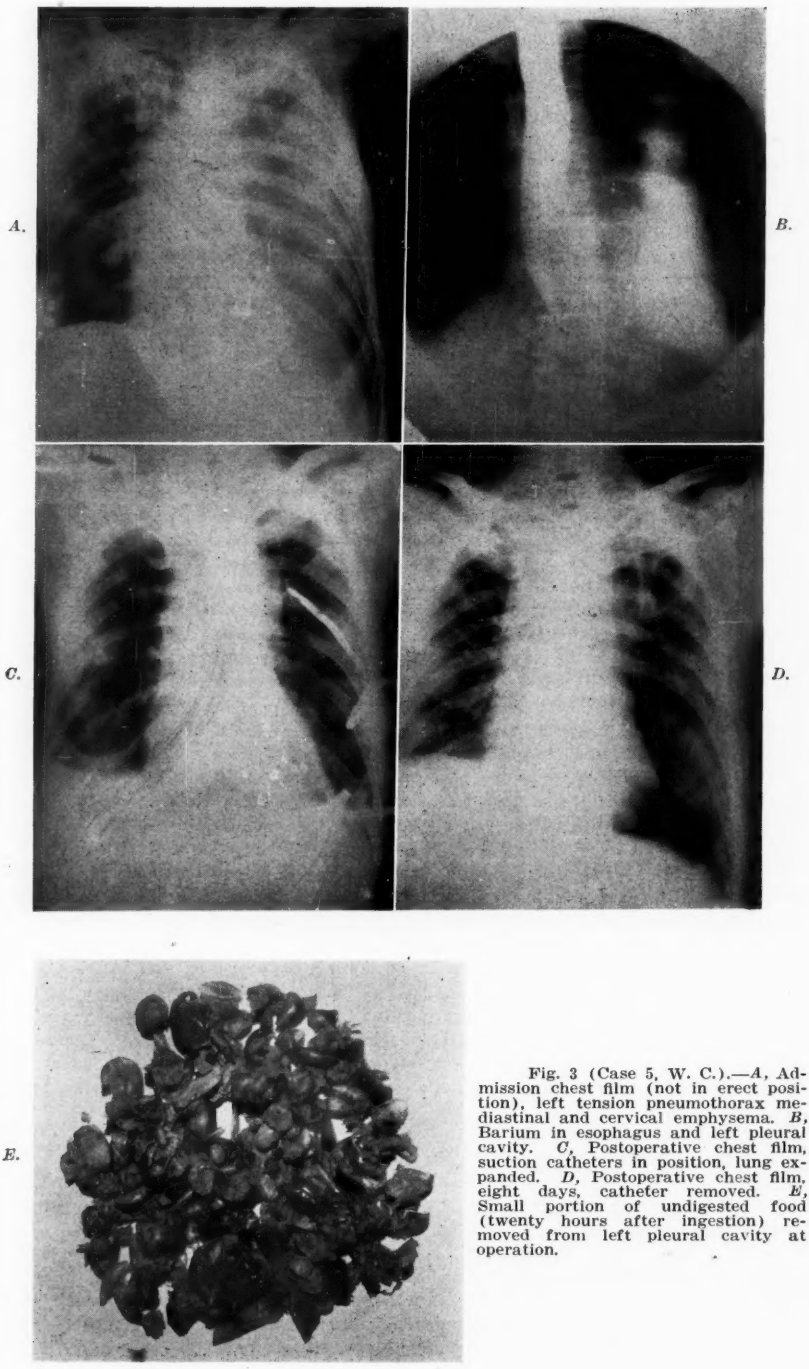

Fig. 3 (Case 5, W. C.).—*A*, Admission chest film (not in erect position), left tension pneumothorax mediastinal and cervical emphysema. *B*, Barium in esophagus and left pleural cavity. *C*, Postoperative chest film, suction catheters in position, lung expanded. *D*, Postoperative chest film, eight days, catheter removed. *E*, Small portion of undigested food (twenty hours after ingestion) removed from left pleural cavity at operation.

Postoperatively the patient was kept in an oxygen tent, and penicillin and sulfadiazine were administered in adequate dosage. Intravenous fluids and blood were administered as indicated. Continuous suction was maintained on the intercostal catheters and a considerable volume of thin serous fluid obtained daily. The lung remained expanded well, and little or no fluid was allowed to collect in the pleural cavity. The postoperative course was somewhat stormy with elevated temperature, and though the blood pressure remained well above 100 most of the time, he had several sinking spells of short duration, the cause of which could not be determined. Continuous nasal suction was maintained for the first six days, following which small amounts of fluid were injected directly through the tube into the stomach.

Fig. 4 (Case 5, W. C.).—Post-mortem specimen; spontaneous rupture of lower esophagus on ninth postoperative day.

A roentgenogram of the chest on February 24, five days postoperatively, revealed the lung fully expanded with no change to the parenchyma of the lung. The general condition improved and everyone was certain that the patient was going to recover. On Feb. 26, 1946, he was alert and cooperative, and apparently had few complaints. X-ray view of the chest on this day, however, showed an area of mottled increased density in the peripheral portion of the right lung field, which was interpreted as a patch of pneumonia. He continued to improve until February 27, eight days postoperatively, when he suddenly complained of severe pain in the right side of the chest, and for a time was irrational. Blood pressure dropped, but after a short time came up again, and apparently he was all right. There was no expectoration of blood, no edema, tenderness, or other findings to suggest a process in the lower extremities. Toward midnight he had another attack of pain in the chest and rapidly went downhill, dying at 6:40 A.M. on Feb. 28, 1946, on the ninth postoperative day.

Post-mortem examination revealed that the immediate cause of death was pulmonary embolism. The main branch to the right pulmonary artery was obstructed by a large antemortem clot, the right lung also containing two small fresh hemorrhagic infarcts, showing that he had had minor embolic episodes prior to the terminal embolus. The source of the embolus was not demonstrated; the leg, veins, and pelvic veins were free of blood clots. The left lung showed partial atelectasis and also a small infarct. The left pleural cavity showed a suppurative pleuritis, but contained little fluid. There were still a few food particles clinging to the surface of the left lung. The esophagus showed a linear area of ulceration on the posterolateral aspect of the wall just above the cardia. This tear measured 4 cm. in length. The outer surface was partially closed by sutures which had partially broken down. There was very little evidence of healing. The surrounding mucosa of the esophagus showed evidence of acute esophagitis with no evidence of any acute or chronic ulcers in the stomach or duodenum. Microscopic examination of the esophagus showed the wall to be necrotic and infiltrated with large numbers of neutrophiles, but with no evidence of malignancy. It is unfortunate that this man died of a pulmonary embolism or that the preliminary signal emboli were not recognized and venous ligation carried out, for with the postoperative course progressing satisfactorily we have every reason to believe that he would have recovered from this serious illness.

Spontaneous perforation of the lower esophagus into the mediastinum or pleural cavity is always a tragic occurrence. It presents a clinical picture so characteristic that the diagnosis should be made in the majority of instances if the possibility of such a condition is considered. While it may be produced by mechanical factors alone, the importance of a local esophagitis weakening the esophageal wall must be recognized. To date the correct diagnosis has been made during life fourteen times[4, 5, 7, 14, 16, 21, 23, 46, 50, 73, 74] including Cases 3, 4, and 5 in the present series. From this group operative repair has been carried out five times, four transpleural[5, 14, 16] with our Case 5 and one transabdominal,[23] with recovery in two patients.[5, 16] Early diagnosis, prompt surgical repair, and proper postoperative treatment may be expected to save a considerable number of these patients from an otherwise certain fatal termination.

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SPONTANEOUS RUPTURE OF THE ESOPHAGUS

Report of One Case, With Recovery

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SPONTANEOUS rupture of the esophagus is a very unusual occurrence. In August, 1946, Eliason and Welty* reviewed forty-one recorded cases of rupture (not perforation) of the esophagus, and added case histories of three of their own patients. They stated that no cases had been reported in the literature in which recovery had taken place, although in an editorial footnote in the 1944 *Year Book of Surgery,* Graham mentioned two cases which resulted in empyema with recovery following drainage.

Since the condition of spontaneous rupture of the esophagus is so rare, recovery so unusual, and the surgical management so complicated, it is hoped that this presentation of a case with recovery will be helpful to one confronted with the same problem.

CASE REPORT

R. E. W., a 33-year-old veteran of World War II, was admitted to the Veterans Administration Hospital, Oteen, N. C., on June 2, 1946, at 11 P.M. and gave the following history:

From 1937 to 1939, the patient complained of irregular attacks of "acid indigestion" and was treated for a supposed peptic ulcer. No radiographic studies were made. From 1939 to 1944 he had occasional upper abdominal discomfort which was relieved by sodium bicarbonate. On one occasion in 1941, he complained of sudden, severe pain in the upper abdomen. He became acutely ill, was hospitalized, operated upon, and recovered. He was told by his surgeon that he had acute pancreatitis. After this episode he felt well with an occasional dose of soda and without dietary restrictions until May, 1946. He served as a master sergeant in an engineers' battalion from May, 1943, to March, 1946. Twelve months of this service were spent in the tropics and during the entire thirty-four months he was not hospitalized.

On May 18, 1946, the patient consumed five highballs from 5 to 10 P.M., but was not intoxicated and did not vomit. He went to bed at 11 P.M. and arose at 8 A.M. He ate breakfast consisting of fruit juice, cereal, eggs, toast, and coffee. He then walked three-fourths of a mile to buy a paper and felt perfectly well. While at the paper stand, however, he was seized with a sudden, severe pain in the left epigastrium, which "doubled him up." He immediately vomited bloody material. He rode home in a cab and vomited three times on the way, the vomitus being described as bloody. The pain and nausea persisted and he was admitted to a hospital in his own community about noon on May 19, 1946.

The surgeon's notes stated that the patient, in obvious pain, was lying in bed with both knees flexed. Physical examination revealed boardlike rigidity of the left rectus muscle and marked rigidity of the right rectus muscle with tenderness across the entire upper abdomen.

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Read (by title) at the Twenty-Seventh Annual Meeting of the American Association for Thoracic Surgery, St. Louis, Mo., May 28, 29, and 30, 1947.

*Eliason, E. L., and Welty, R. F.: Surg., Gynec. & Obst. 83: 234, 1946.

The leucocyte count on admission was 6,400, but rose in a few hours to 9,100. The temperature was 99° F., pulse rate 100, and respirations 26 per minute. On admission a roentgenogram (Fig. 1) was made and was interpreted as negative. A tentative diagnosis of perforated peptic ulcer was made and a laparotomy was performed at 5:30 P.M., about 5 hours after admission. No perforation was found and a careful search of the peritoneal cavity revealed no pathology to account for the symptoms. The omental tissue had a peculiar appearance and a piece was removed for examination before the abdominal cavity was closed. The pathologist's report was fibrosis of the omentum, probably due to an old pancreatitis.

Fig. 1.—Roentgenogram made before operation on May 19, 1946. Note area of lessened density behind the heart. In the light of the subsequent history we feel that this shadow may be characteristic of the condition described.

The postoperative period was stormy. The temperature rose to 103.4° F.; the pulse rate increased to 120, and respirations to 34 per minute shortly after operation. The next day, May 20, crepitant râles were heard and later that day a friction rub was noted over the left lower lobe. On May 21, the left lower part of the chest was tympanitic and there was some dyspnea. On May 25, a roentgenogram of the chest (Fig 2) was made, which showed a wide mediastinum and a left hydropneumothorax. Thoracentesis yielded 135 c.c. of clear, amber fluid which was negative on smear for organisms. The cells in the fluid were predominantly lymphocytic. On May 28, the thoracentesis was repeated and 240 c.c. of air withdrawn. No fluid was obtained, but the procedure relieved the dyspnea. During the next four days several thoracenteses were performed, but only small amounts of fluid and air could be obtained.

During this period, the temperature was septic and ranged from 101 to 106° F.; the pulse rate varied from 90 to 120; and the respirations from 24 to 40 per minute. He was given 1,940,000 units of penicillin intramuscularly from May 20 to June 2. He took some liquids by mouth and was given glucose in saline solution intravenously almost daily.

On June 2, 1946, he was seen by one of us (J. A. M.) in consultation. A thoracentesis by this consultant in the left ninth interspace near the spine yielded several ounces of a brownish, purulent, sour-smelling fluid with acid reaction. It was very suggestive of gastric contents. The next procedure was to give barium by mouth. Under the fluoroscope this was seen to pass rapidly into the stomach. Then a roentgenogram was made with the patient in the upright position; in this film the stomach appeared normal. Next, a film made (Fig. 3) with the patient in the head-down position showed the stomach to be in the left part of the chest. A tentative diagnosis of a diaphragmatic hernia with a perforated ulcer in the herniated portion of the stomach was made.

Fig. 2.—Portable roentgenogram made on May 25, 1946, shows a pneumothorax with some fluid and a wide mediastinum.

The patient was transferred to the Veterans Administration Hospital at Oteen, N. C. On June 3, 1946, the red blood cell count was 3,340,000, hemoglobin 9 Gm., and the leucocyte count was 10,900 with 80 per cent neutrophiles. The sedimentation rate was 31 mm. per hour. The urine showed a trace of albumin with an occasional white and red blood cell and a few cylindrical casts. The blood pressure was 125/60. Moist râles were present over the left upper lobe and there were diminished breath sounds at the base. X-ray examination showed the left lung to be partially collapsed with a broad adhesion between the first and second ribs. There was a collection of air medially and laterally in the upper third of the pleural cavity and of fluid and air just above the diaphragm. There was a streak of barium above the left border of the heart outside of the gastrointestinal tract.

On the assumption that the patient had empyema as a result of a perforation in the herniated portion of the stomach, a left thoracotomy was done at the level of the ninth rib in the posterior axillary line, under local anesthesia. After resection of 4 cm. of the rib, a large drainage tube was inserted and fixed in place with a safety pin and adhesive dressing.

Culture of the fluid removed at operation showed *Staphylococcus areus,* nonhemolytic streptococci, and a yeast form identified as Monila. No acid-fast bacilli were found.

Penicillin was given at the rate of 20,000 units every three hours. A Levine tube was passed intranasally into the stomach and suction applied. Part of the water or milk given by mouth escaped through the thoracotomy drainage tube. A tablet of methylene blue was given orally and the blue dye appeared in a short time in the drainage through the thoracotomy tube. Nutrition was maintained by the intravenous administration of glucose in saline and amigen solution.

On June 6, 1946, under spinal anesthesia, a jejunostomy by the Wangensteen technique with a 14 French catheter was done by one of us (J. A. M.). After this, the patient was given nothing by mouth and was fed approximately 3,000 calories a day, supplemented by vitamins A, B, D, and ascorbic acid through the jejunostomy tube. This was supplemented by several intravenous infusions of glucose in saline and amigen and by four blood transfusions of 500 c.c. each of citrated blood between June 6 and June 28. The thoracic cavity was irrigated four times daily with a 1 per cent sodium bicarbonate solution.

Fig. 3.—Roentgenogram made June 2, 1946, in the head-down position after ingestion of barium sulfate. This was interpreted as being a diaphragmatic hernia.

During the first few days, there was a reversal of the albumin-globulin ratio, but the total protein, nonprotein nitrogen, chlorides, and blood calcium remained within normal limits. The temperature in the meantime had gradually subsided and the drainage from the left side of the chest gradually ceased.

By June 28, twenty-two days after jejunostomy, the patient's condition had so improved that we felt he could undergo the major procedure of repair of the supposed diaphragmatic hernia. Under intratracheal anesthesia of nitrous oxide, oxygen, and ether, supplemented with Pentothal sodium during induction, the operation was undertaken by both of us (J. A. M. and J. D. M.). The incision was made over the left ninth rib with resection of the ninth rib anterior and posterior to the thoracotomy wound. The eighth rib was next divided behind the angle. The pleura was incised in the bed of the ninth rib and a rib spreader inserted.

The left lung was found adherent to the diaphragm and to the lateral chest wall. There was an anterior empyema space and a posterior space which communicated with the fistula. The lung was separated from the diaphragm with difficulty but without injury. After the lower lobe of the lung had been completely freed, a large aperture was palpated in the diaphragm at the site of the esophageal hiatus. This appeared to be large enough to permit a hernia, but the stomach was not present in the thoracic cavity.

The left diaphragm was then incised, the abdominal cavity entered, and the stomach identified. The incision in the diaphragm was extended to the hiatus and the esophagus visualized. A rent 4 cm. long was found in the anterolateral portion of the esophagus just above the diaphragm. This was unquestionably an old tear, because the mucous membrane was everted and healed to the muscular coat. The mucous membrane itself appeared perfectly normal. The tear extended to the cardia. At the upper end of the tear the esophagus was fibrous and adherent to the aorta at that point. No satisfactory explanation for this lesion could be arrived at. The lower end of the esophagus was separated from the aorta and was completely mobilized.

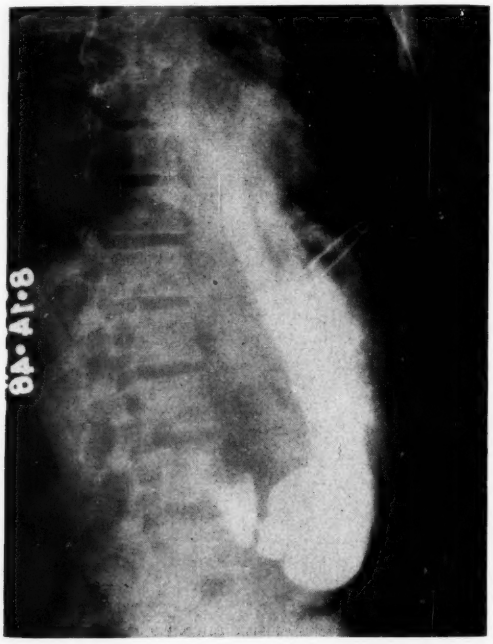

Fig. 4.—Roentgenogram made on Aug. 2, 1946, in the head-down position after ingestion of barium shows the stomach to be in the abdomen.

An incision about 4 cm. long was made through the cardia and into the fundus of the stomach. An esophagogastroplasty was then done approximating the edge of the esophagus to the cut surfaces of the stomach with interrupted Connell sutures of silk. This was reinforced with a continuous silk suture. The incision in the diaphragm was closed with interrupted mattres sutures of silk down to the hiatus. The fundus of the stomach was sutured to the diaphragm to remove tension from the line of repair. A large rubber tube for drainage was inserted at the site of the original thoracotomy and the wound was closed in layers around it.

Convalescence was remarkably smooth. Feeding was continued through the jejunostomy tube until the tenth postoperative day, at which time the tube was removed. No leakage

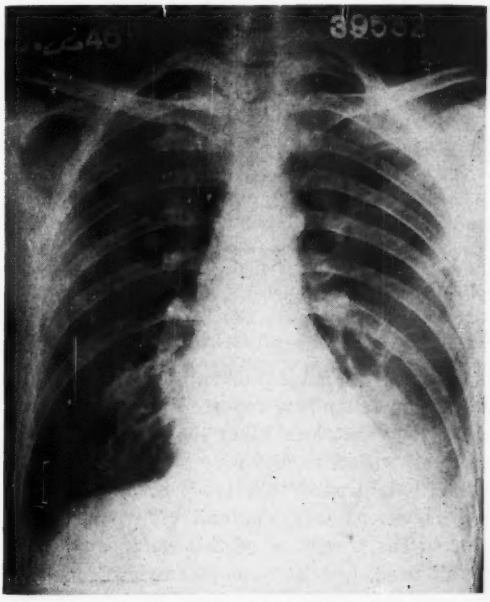

Fig. 5.—Roentgenogram made Oct. 22, 1946, showing lung to be completely re-expanded.

Fig. 6.—Roentgenogram after ingestion of barium sulfate, showing esophagus to be normal.

occurred. Liquids by mouth were started in small amounts on the seventh postoperative day, and on the fourteenth day a high caloric, bland diet was given. A regular diet was instituted on August 12 and the patient has experienced no difficulty in swallowing at any time.

Following the latter operation, the expected rise in temperature was moderate and of short duration and it has been entirely normal since July 23. The drainage from the pleural cavity gradually decreased as the left lung re-expanded. The drainage tube was removed on Sept. 21, 1946. The wound has completely healed. Penicillin was continued until August 1, a total dosage of 9,120,000 units having been given. The patient was allowed out of bed on Aug. 2, 1946, and was gradually allowed to increase activities. The stomach is in the abdomen (Fig. 4), as shown by the roentgenogram taken in the head-down position. The lung was completely re-expanded (Fig. 5) and the esophagus appeared normal (Fig. 6). He was discharged on Oct. 24, 1946, completely recovered.

COMMENT

In their discussion of the subject of rupture of the esophagus, Eliason and Welty stated: "The mortality rate reported so far in spontaneous rupture of the esophagus is 100 per cent, excluding the two cases mentioned by Graham. Most patients succumb within twenty-four hours, though one patient survived seventeen days. With improved techniques in anesthesia, freer use of blood and plasma for intravenous infusion, and effective chemotherapeutic agents, the limiting factor in the treatment of this condition is that of diagnosis. It does not seem at all unlikely that soon recoveries from this condition will be reported."

Spontaneous rupture of the esophagus should be considered in the differential diagnosis in any patient who suffers severe, upper abdominal pain associated with vomiting, particularly of blood. If this syndrome is accompanied by cervical emphysema or the development of a pneumothorax, the lesion should be strongly suspected. We think that when a pocket of gas is discovered behind the heart, as shown in Fig. 1, we would immediately make the diagnosis. Two other procedures should be carried out to confirm the diagnosis: first, the administration of barium with subsequent roentgen films. The presence of barium in the chest would confirm the diagnosis. Second, if on thoracentesis, gastric contents were aspirated, this would indicate either a rupture of the esophagus or a perforation of the herniated stomach.

As soon as the diagnosis is made, drainage of the thoracic cavity should be instituted. No food or drink should be given by mouth and a jejunostomy should be done in order to feed the patient. Antibiotic agents should be given in adequate doses. Nutrition must be maintained. Glucose and amino acids, blood or plasma transfusions are indicated to supplement jejunal feedings. We believe that with early recognition of the condition and prompt application of the measures outlined here, many of these patients can be saved. In all probability, the rupture would heal spontaneously and if it did not, repair could be successfully effected at a later date. We question the wisdom of immediate repair of the ruptured esophagus in those patients whose desperate condition following the accident is a strong contraindication to any major surgical procedure.

DIVERTICULA OF THE THORACIC ESOPHAGUS

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SINCE Zenker's classification, in 1877, of esophageal diverticula into pulsion and traction types, many individual case reports and a few larger series of diverticula of the thoracic esophagus have been reported. A much larger series of diverticula of the pulsion type in the pharyngocervical esophagus has been reported, and the surgical management, technical aspects, and the treatment of these cases have become well established. In addition, these reports have described fully the pathologic and clinical aspects of these two now well-accepted and well-established types. Diverticula of the thoracic portion of the esophagus, however, are far less common and very little has been written regarding their specific surgical management. I wish, therefore, to present a small series of diverticula of the thoracic esophagus in which the patients required surgical relief of their symptoms, and to describe in more detail their surgical management and the technical aspects of these operations.

The clinical course and symptomatology are quite characteristic. The symptoms are related to a progressive mechanical obstruction of the esophagus and range from indefinite gastric symptoms, substernal distress and pain, varying degrees of dysphagia, regurgitation, and vomiting. Depending, of course, on the severity of these symptoms, there are an associated debility and malnutrition reaching severe states in patients in whom the mechanical obstruction has become practically complete. These severe symptoms and severe stages of malnutrition are chiefly associated with the large pulsion diverticula and rarely with the traction type. The traction type is most commonly seen in the midesophageal region associated with the hilar regions of the lungs and the various inflammatory conditions involving the lungs and the hilar nodes. They are produced by an extension of an adjacent inflammatory reaction to the wall of the esophagus, with traction and outpocketing of the entire thickness of the wall of the esophagus as the adjacent acute inflammatory reaction subsides and is replaced by scar and scar retraction. In this type the sac rarely hangs down in a dependent position in relation to the esophagus or is rarely large enough to angulate or obstruct the esophagus and cause the classical symptoms. Surgical intervention, therefore, is not indicated except for other complications primarily caused by the associated infection—periesophageal abscess or bronchial fistula. These complications, however, are much more commonly associated with carcinoma which, with considerable frequency, produces pseudodiverticula or esophagotracheal or esophagobronchial fistulas. If carcinoma can be ruled out by bronchoscopy and esophagoscopy and properly taken biopsy specimens, the surgical

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repair and excision of such fistulas are indicated. Such pseudodiverticula and fistulas (Fig. 1) when associated with carcinoma definitely indicate its inoperability.

The rare case of esophagotracheal fistula resulting from a traumatic or inflammatory process can be cured by operation and this must be accomplished to save the patient's life. A transpleural posterolateral approach, ligating and sectioning the azygos major vein with excellent mobilization and visualization of the membranous portion of the trachea and esophagus as well as the fistulous tract (Fig. 2) will permit a meticulous excision of the fistula or closure of the trachea and esophagus and, if massive doses of penicillin and streptomycin are used, primary healing without residual infection can be expected.

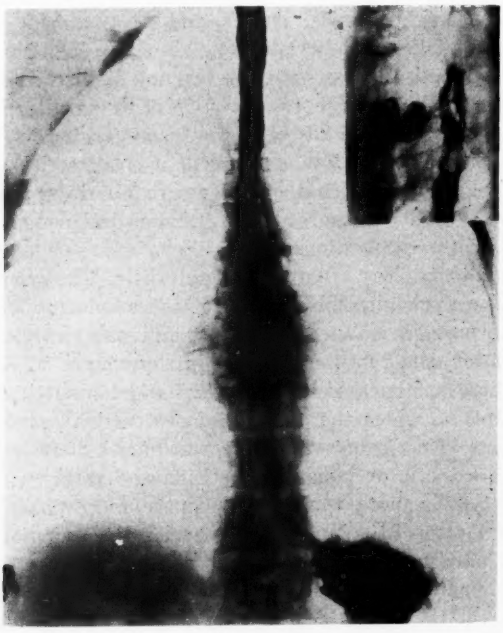

Fig. 1.

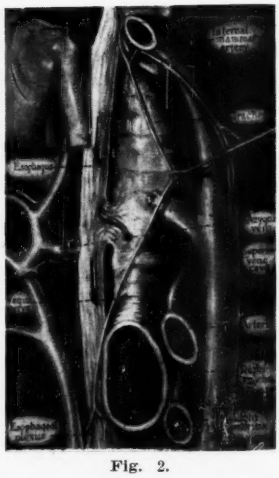

Fig. 2.

Fig. 1.—Barium study of esophagus, showing perforation of a carcinoma of the esophagus into the mediastinum and right lung, producing a pseudodiverticulum.

Fig. 2.—Operative sketch showing the exposure obtained through the right thorax of the esophagotracheal region for repair of esophagotracheal fistula.

The pulsion type of thoracic diverticula, however, is similar clinically to those so frequently seen in the esophagopharyngeal region. These diverticula produce symptoms and nutritional decline of the patient owing to the dependent position of the sac which fills with liquid and food and gradually becomes large enough to produce mechanical angulation and obstruction of the esophagus. When the diverticulum reaches this stage of obstruction, it seriously jeopardizes the patient's health and life, and only surgical treatment can be expected to give him relief of these serious symptoms. Although a

large number of pulsion diverticula of the thoracic esophagus has been reported in the literature, there are very few references to specific surgical treatment of this lesion. Esophagoscopy, dilatation, sounding, lavage, and dietary management have been described, which, however, cannot be successful in the large, truly obstructing diverticula. Exclusive of those associated with diaphragmatic hernia, five patients have been operated upon at The Lahey Clinic who had large pulsion diverticula of the thoracic esophagus with marked symptoms of obstruction. Three of these diverticula occurred before the advent of penicillin and the patients were extremely bad risks because of extensive cardiovascular disease. In these cases a transthoracic diverticulopexy was done, with the patients' survival and complete relief of symptoms for their remaining years of life.

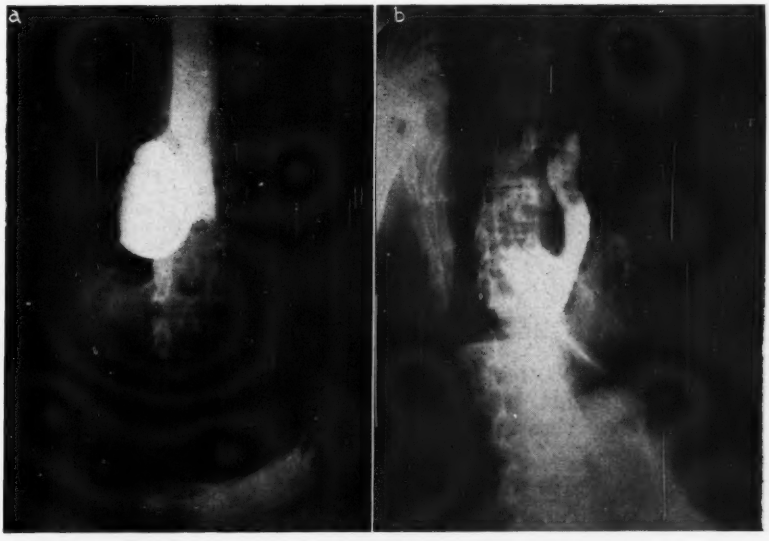

Fig. 3.—Barium study of the esophagus showing a large pulsion diverticulum of the lower thoracic esophagus. *a*, Preoperatively, the sac in a dependent obstructing position; *b*, postoperatively, showing adequate drainage and relief of obstruction following diverticulopexy.

REPORT OF CASES

CASE 1.—A man, 49 years of age, was admitted to the clinic Dec. 8, 1936, complaining of anorexia, dysphagia, vomiting, and progressive weight loss. Roentgenograms showed a large pulsion diverticulum, about 5 by 5 cm., of the lower third of the esophagus (Fig. 3, *a* and *b*). He was admitted to the hospital, and on Jan. 2, 1937, transthoracic diverticulopexy was done. Following this he made a good recovery and was completely relieved of the esophageal symptoms. He was readmitted to the hospital approximately one year later, however, with dyspnea, cough, severe hemoptysis, and fever, and died approximately one week after admission.

Post-mortem examination showed extensive miliary tuberculosis as the cause of death. The operative field was in satisfactory condition.

CASE 2.—A 61-year-old man was admitted to the clinic Jan. 18, 1940, complaining of pressure in the chest, dysphagia, and regurgitation of food; the symptoms had been progressive

over a period of two years. Roentgenograms showed that the upper two-thirds of the esophagus was dilated; a large diverticulum, 8 by 8 cm., was present at the junction of the lower and middle thirds of the esophagus, with complete obstruction.

The patient was admitted to the hospital on Jan. 19, 1940, and on January 29, a transpleural diverticulopexy was done. He made an uneventful convalescence and was completely relieved of the symptoms. He returned to the clinic two years later with definite angina pectoris but no recurrence of difficulty in swallowing or regurgitation. Diagnosis was made of coronary sclerosis, and restricted activity and a medical regimen were advised.

The patient died in September, 1945, of coronary occlusion, about five years after operation for the diverticulum, without recurrence of the esophageal symptoms.

CASE 3.—A 72-year-old man was admitted to the clinic Jan. 15, 1940, complaining of retrosternal distress, dysphagia, regurgitation, and progressive loss of weight and strength of about three years' duration. Roentgenograms showed a large diverticulum in the lower third of the esophagus. The patient was in relatively poor general condition from extensive general arteriosclerosis and associated angina pectoris and pulmonary emphysema. In view of the serious esophageal symptoms, however, he was admitted to the hospital Jan. 15, 1940. On Jan. 19, 1940, a transpleural diverticulopexy was done, with complete relief of the esophageal symptoms until the present time.

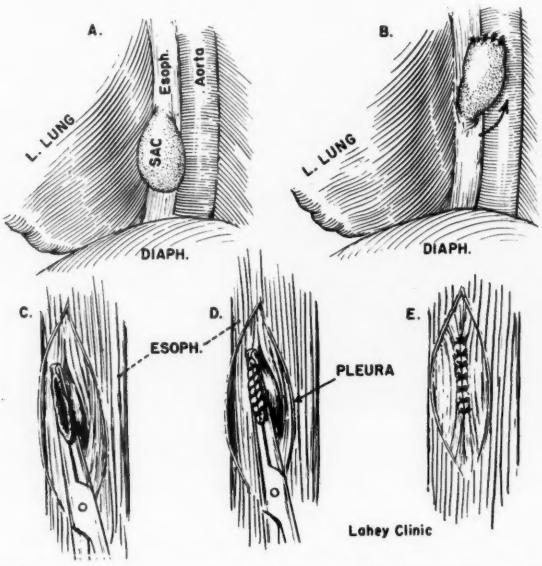

Fig. 4.—Sketches of the operative procedures for the treatment of thoracic diverticula. *A*, Sac in its usual dependent relation to the esophagus; *B*, elevation and fixation of the sac in diverticulopexy; *C*, *D*, and *E*, operative steps in one-stage excision of the sac.

In these three cases transpleural exposure was made through the left lower thorax, the mediastinal pleura overlying the diverticulum was opened, and the diverticulum and esophagus adjacent to the neck were mobilized. The apex of the sac was elevated along the long axis of the esophagus and sutured with fine silk to the pleura at the highest possible point of elevation which would not produce traction or any angulation of the esophagus itself (Fig. 4, A and B). This permitted free drainage of the sac at all times and prevented the

food from passing into the diverticulum, rather than into the esophagus proper, during deglutition. This surgical procedure proved to be a satisfactory method of managing these three cases of intrathoracic diverticula before the advent of penicillin. The danger of mediastinal and pleural contamination that would have been associated at that time with any attempt to excise the diverticulum would have been great and the extremely poor general status of these patients would have made a more prolonged procedure dangerous. The patients were completely relieved of symptoms for the duration of their lives (from one to six years).

In the past year, however, two younger patients have been seen who required operative treatment for relief of the symptoms of an obstructing thoracic esophageal diverticulum. In these cases the diverticulum was excised in one stage, in conjunction with intensive treatment with penicillin.

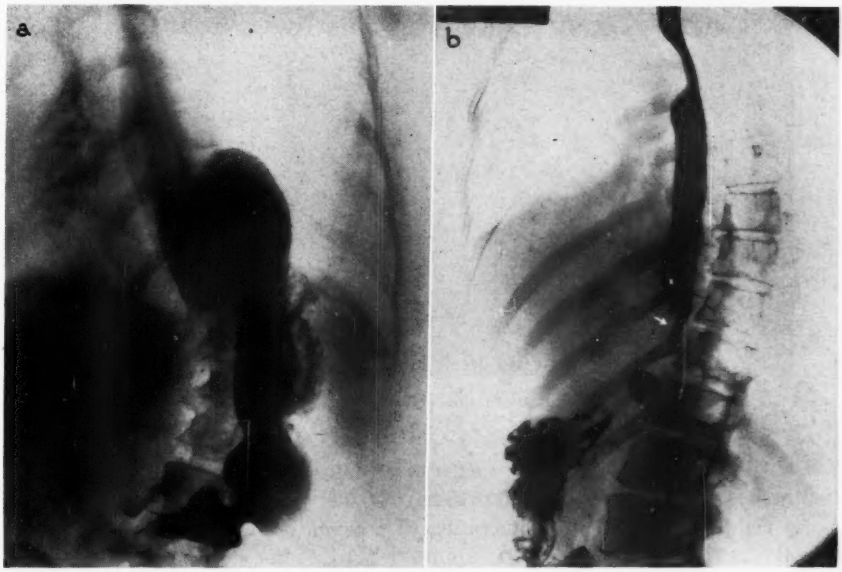

Fig 5.—Barium study of large obstructing pulsion diverticulum; *a*, preoperative appearance of the sac; *b*, appearance after excision.

CASE 4.—A woman, aged 47 years, was admitted to the clinic April 24, 1946, complaining of progressive dysphagia, regurgitation, vomiting, and loss of weight and strength over a period of ten years. Roentgenograms showed a large diverticulum of the lower third of the esophagus (Fig. 5, *a* and *b*).

The patient was admitted to the hospital on June 5, 1946. On June 7 a transpleural excision of the diverticulum in one stage was done. Following this procedure she made a very satisfactory recovery, with complete relief of the symptoms.

CASE 5.—A man, aged 53 years, was admitted to the clinic in August, 1946, complaining of regurgitation of food and retrosternal distress of about six months' duration. In addition, he had noted marked dyspnea, palpitation, tachycardia, nervousness, and weight loss.

Roentgenograms showed a very large diverticulum at the junction of the lower and middle thirds of the esophagus. He also had the clinical signs and symptoms of hyperthyroidism and the basal metabolic rate was plus 45. A diagnosis was made of primary hyperthyroidism and diverticulum of the thoracic esophagus (Fig. 6, *a* and *b*). He was placed on propylthiouracil and after proper preparation, he was readmitted in November, 1946. On Nov. 13, 1946, subtotal thyroidectomy was done.

Since at this time the hyperthyroidism was fully controlled, on November 26 a transthoracic excision of the esophageal diverticulum, one stage, was done. Convalescence was satisfactory and uneventful, and the patient had complete relief of the symptoms.

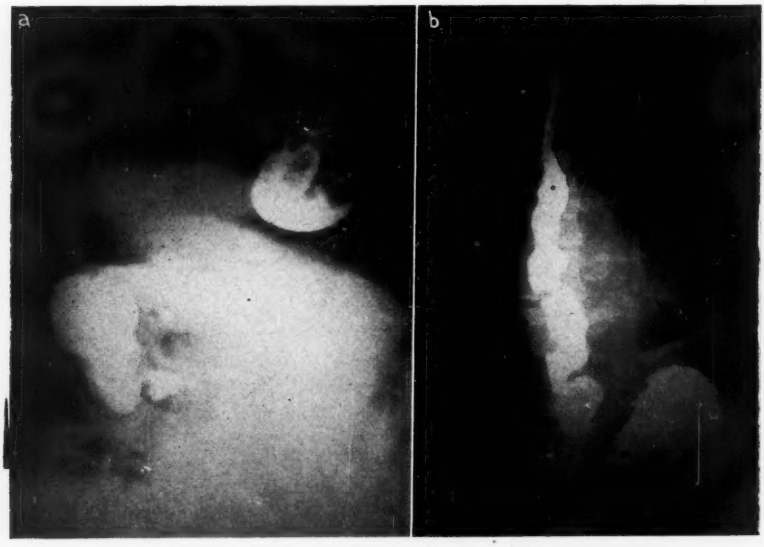

Fig. 6.—Barium study of a large obstructing diverticulum lying transversely behind the heart; *a*, preoperative roentgenogram; *b*, appearance after excision.

In these two cases the diet was restricted to high-caloric liquids, and penicillin was administered intramuscularly, 30,000 units every three hours for forty-eight hours prior to the operation. The operation was carried out through a wide transpleural posterolateral approach through the left lower thorax.

In Case 4 the diverticulum could be visualized lying immediately beneath the mediastinal pleura. The pleura was opened and the sac carefully and completely mobilized, dissecting back the thinned-out muscle fibers from around the neck of the diverticulum.

In Case 5, upon opening the thorax, the diverticulum could not be visualized since it lay transversely behind the pericardium. It became necessary to mobilize the lower third of the esophagus and to dissect this sac out carefully from the mediastinum. In this case as well, the muscle fragments were carefully dissected from the neck. A thin Kocher clamp was then placed across the neck into the sac about 0.5 cm. from the wall to allow sufficient tissue so that it might be turned in without encroaching on the lumen of the esophagus

itself. A second Kocher clamp was placed adjacent to this and the diverticulum removed between these clamps, after carefully walling off the entire pleura and surrounding tissues with cellophane-lined gauze pads (Fig. 4, C, D, and E). The mucosal layer contained in the thin Kocher clamp was then closed by a running over-and-over suture of fine chromic catgut, and the mucosa closed by drawing up on this running suture as the Kocher clamp was removed, thereby accomplishing a relatively aseptic closure of the mucosal layer. The muscular layers of the esophagus were then closed with fine interrupted silk sutures. The esophagus was then returned to its bed in the mediastinum, the mediastinal tissues were sprayed with 100,000 units of penicillin solution, and the mediastinal pleura was closed. Another 100,000 units of penicillin were sprayed throughout the pleural cavity, the lung re-expanded, and the chest and the thorax closed, leaving an intercostal catheter for suction to aid in rapid re-expansion of the lung. The patient was placed on 40,000 units of penicillin intramuscularly for ten days following operation, and the intake by mouth was restricted and graduated much in the same manner as in the management of a patient who has had a gastric resection.

By this technique convalescence in these cases was uneventful without complication or residual infection. All of these patients obtained complete and lasting relief from the esophageal symptoms. Although there may still be an occasional specific indication for the use of the diverticulopexy type of operation, it is recommended that, with proper use of penicillin, transpleural excision of the diverticulum by the described technique, in one stage, is the operation of choice.* This operation is recommended and should be carried out on all patients suffering from symptoms of esophageal obstruction which arises from a diverticulum of the thoracic esophagus.

*Since this paper was submitted for publication, three additional patients have been operated upon by this method.

TUBERCULOSIS AND PREGNANCY

An Analysis of One Hundred Eighteen Cases

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ABOUT a decade ago one of us[1] published a report of six cases of pulmonary tuberculosis complicated by pregnancy in which the delivery was by cesarean section. All of these patients had artificial pneumothorax at the time of delivery. At that time we felt that the tuberculous woman nearly always improved when she became pregnant unless there was some other complication. We felt that there was some factor in the delivery that caused the patient's tuberculosis to become worse after the baby was born. We did not know what this factor was. Since then, we have decided that the cause of the flare-up was the greatly increased intrapulmonary pressure that developed during the second stage of labor. It seems clear that incompletely healed tuberculous foci would be torn apart by this pressure, and reactivation of the inactive lesions would result.

Friedman and Garber[2] in an excellent review of the available and pertinent literature on the subject have pointed out the wide difference of opinion as to the effect that pregnancy has on tuberculosis. In reviewing the literature, we have classified the articles into seven groups as follows: Group I,[3-36] which believed that pregnancy was either beneficial or harmless; Group II,[37-72, 160] which believed that pregnancy was harmful; Group III,[73-94] which believed the patient was better during pregnancy but became worse after delivery; Group IV,[95-116] which believed delivery was harmful; Group V,[117-121] which believed lactation produced the harmful effects; Group VI,[122-136] which believed the type of disease (caseous pneumonic, exudative, or proliferative) was the chief deciding factor; and finally, Group VII, [137-156, 160] which believed that collapse therapy enhanced the chances of a favorable outcome in patients with tuberculosis and pregnancy.

In Group I the reasons given for the improvement were: "(a) Mother fixes more nitrogen[7]; (b) She utilizes in a better manner the sulphur of her albumenoids and the iron of her food and at the same time mobilizes a larger quantity of phosphorus and lime; (c) more oxygen is consumed and more carbon dioxide produced. All of this forms a hyper nutrition which is beneficial to the mother;" (d) increased intra-abdominal pressure acts on the lung like phrenic avulsion or artificial pneumothorax, [23, 24, 25] or pneumoperitoneum[26]; (e) presence of gonadotrophic hormone in pregnant women[27] and (f) "all the physiological

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processes in the pregnant woman are functioning at their best during this time.[29]''

In Group II the reasons given for the harmful effects of pregnancy were: (a) Increased demands on metabolism, oxygenation and innervation, on circulation and elimination[38, 39, 41, 45, 50, 66, 67, 68]; (b) point of least resistance is put to test; (c) presence of the gravid uterus interferes with respiration and aeration of blood; (d) nausea and vomiting of pregnancy tend to interfere with assimilation; (e) prolonged muscular exertion of labor, loss of blood and attending exhaustion, use of anesthetics, auto infection by the aspiration of infected material from an old focus in the lung into healthy portions, and the possibility of toxins thrown into the system from the placental site; (f) strain of subsequent lactation and responsibility incident to the care of the child; (g) hepatic insufficiency[42]; (h) ''pregnancy seems to reduce the natural defense forces[47];'' (i) ''the depressive and damaging action on the adrenals and other glands[51];'' (j) ''upward displacement of the diaphragm[53];'' (k) ''decreased lipolytic activity of the blood causes difficulty in dissolving fatty capsules of bacilli;'' (l) ''increased cholesterol content of the blood on which tubercle bacilli thrive;'' (m) decalcification liberates bacilli; (n) increased basal metabolic rate[55]; (o) renal adequacy declines; (p) reduced tuberculin reactivity[56]; (q) respiratory rate increased; (r) alteration in the permeability of the capillaries; (s) contraction of the thorax[61]; (t) tissue changes in the lungs and air passages; (u) strength of woman is diminished[65]; (v) ''the blood of the pregnant woman is full of lipoids which favor the growth of tubercle bacilli and anatomical changes in the mucous membranes of the body, especially the larynx and bronchi which favor the development of tuberculosis.[72]''

In Group III the explanations of the patients' getting worse after delivery following pregnancy during which their condition improved were as follows: (a) ''A great tendency to hemorrhage during puerperium[73];'' (b) ''straining and increased blood pressure incident to labor probably converts partially healed lesions into open ones, thus liberating tubercle bacilli in large numbers[74];'' ''many free organisms are thrown into the blood stream[3]; actual physiologic exhaustion[3], the congestion of the lungs incident to labor; possible loss of blood; and effects of anaesthetic (if one has been used;'' (c) ''cardiac failure and pulmonary edema[76];'' (d) ''abrupt displacement of the diaphragm coincident with the emptying of the uterus[77, 82, 85-88];'' (e) ''the change in intrathoracic mechanics during pregnancy accounts for the improvement in the pulmonary condition during the latter months of pregnancy and ill effects which are often noted after parturition.[83]''

In Group IV the reasons given for the harmful effects of delivery were: (a) ''Cardiac failure and pulmonary edema and hemorrhage[95];'' (b) ''*straining at labor may break down practically healed lesions,*[96]'' ''actual physiological exhaustion following labor;'' (c) ''increased capillary permeability which is known to occur at this time.''[98]

Group V believed that lactation is dangerous to both mother and child and should not be permitted.[117-121]

In Group VI the opinion was almost unanimous that the most important factor in the harmful effects of pregnancy and the puerperium is exudative and caseous tuberculosis, except in the far advanced with obviously poor prognosis.[122-136]

There is ample evidence[137-156] that pregnant women whose lungs are successfully collapsed stand spontaneous delivery well in most cases. It seems that those with pneumothorax stand delivery better than those with thoracoplasties. Scadding[152] reported spontaneous delivery in a woman with oleothorax without harmful effect.

Our experience has led us to agree with Group III which believed the patient was better during pregnancy but became worse after delivery. The only serious effort to establish the cause of improvement of the tuberculosis during pregnancy is the work of Steinbach and Klein.[27] They showed that the gonadotropic hormone which appears in pregnancy was responsible for increased resistance to tuberculosis in rabbits and guinea pigs. It seems probable that there are other factors involved in this improvement, but no reasonable evidence of such factors has been produced.

In our opinion DeLee, as quoted by Funk,[96] and Jameson[103] came nearer to the real cause of the harmful effects of pregnancy and the puerperium on tuberculosis than anyone else. DeLee said "straining at labor may break down practically healed lesions." Jameson[103] pointed out "the importance of the reduction of voluntary effort in the second stage of labor which results in increased intrapulmonary pressure." Realizing that the tuberculous disease involves mainly the elastic tissue portions of the lungs, we feel that it is obvious that the greatly increased intrapulmonary pressure which develops during the second stage of labor is most likely to tear open partially healed tuberculous lesions. So far as we know, the intrapulmonary pressure during the second stage of labor has never been measured. It has been demonstrated, however, that the intranasal pressure during[158] forced expiration may reach as high as plus 87 to 100 mm.[157] of mercury. We believe it is probable that the intrapulmonary pressure during the second stage of labor is equal to those quoted and possibly even greater at times. This pressure is great enough to tear open partially healed tuberculous lesions.

With these facts in mind, it occurred to us that the harmful effects of pregnancy and the puerperium could be avoided by eliminating the increased intrapulmonary pressure that develops during labor, particularly the second stage. The most obvious way to do this was delivery by cesarean section. This procedure was first suggested by Dr. Charles Sidney Burwell, who recommended it in the case of a tuberculous woman in active labor who at the same time was having a severe pulmonary hemorrhage. It worked satisfactorily in this case, and we felt that it could be used to advantage in selected cases to avoid the harmful effects of delivery.

We later decided that in suitable cases, the delivery might be allowed to progress to the second stage, and then forceps used after the dilatation was complete, and more recently we have employed continuous caudal or continuous spinal analgesia with the final extraction of the baby with forceps.

The purpose of our study was to see if these methods of delivery were satisfactory, and if they had any harmful effect on the progress of the pulmonary tuberculosis, and to see if one method was more satisfactory than another. It also was our purpose to show that therapeutic abortions, because of pulmonary tuberculosis, were rarely justified. We further were impressed by the assertion[125, 126, 128] that only exudative and caseous pneumonic tuberculosis did badly with pregnancy and we wanted to see if our experience coincided with this. We hoped to be able to establish indications for the different methods of delivery; and finally, we hoped to be able to accumulate data on the effect of the greatly increased intrapulmonary pressure on pulmonary tuberculosis.

The material studied included 118 patients, divided into eight groups as follows: (1) cesarean section with collapse; (2) cesarean section without collapse; (3) forceps delivery with collapse; (4) forceps delivery without collapse; (5) spontaneous delivery with collapse; (6) spontaneous delivery without collapse; (7) therapeutic interruption of pregnancy, hysterotomy; and (8) spontaneous abortion.

CRITERIA OF SATISFACTORY POST-PARTUM AND POSTOPERATIVE PROGRESS

The criteria of satisfactory post-partum and postoperative progress were: (1) no increase in temperature or pulse rate (except immediately postoperative or post partum) in three months after delivery; (2) no reversal of negative sputum to positive in three months after delivery; (3) no nonpulmonary tuberculous complications in three months after delivery; (4) no x-ray evidence of progression of the tuberculous lesion in three months after delivery.

There were 34 patients in Group 1 (Table I); 25 had artificial pneumothorax, and 1 of them had bilateral pneumothorax; 22 of these, including the one with bilateral pneumothorax, had satisfactory postoperative courses. Three of

TABLE I. GROUP 1

CESAREAN SECTION WITH COLLAPSE							
	NO. OF CASES	POST-OPERATIVE COURSE SATISFACTORY	POST-OPERATIVE COURSE WORSE	MATERNAL DEATHS FROM DELIVERY	NOT ACCOUNTED FOR	DIED LATER	PROBABLY DIED LATER
Artificial pneumothorax	25	22	3	0	1*	2†	0
Expanded	2	1	1	0	0	0	1‡
Thoracoplasty	3	3	0	0	0	0	0
Phrenic	4	1	3	0	0	3§	
Totals	34	27	7	0	1	5	1

*The one not accounted for had negative sputum, but had pleural effusion that contained tubercle bacilli and the postoperative course was stormy. No death record after three years.

†One died of massive pulmonary hemorrhage five days after operation. The other died of tuberculosis thirty days after operation. Both had pneumothorax for a few days only (five to thirty days) to control hemorrhage.

‡Lung re-expanded because collapse was unsatisfactory and the other lung had far-advanced disease; sputum was positive.

§Two of the three had far-advanced bilateral disease with positive sputum and hopeless prognosis; one of them died three months postoperatively of tuberculous meningitis; the other died of tuberculosis nine months after operation. The third one had moderately advanced disease with positive sputum and died of tuberculosis twenty-seven months after operation.

this group were worse postoperatively. Of the 3 that were worse, 2 had active, bilateral, far-advanced disease with positive sputum, and poor prognosis. One of these two died five days after delivery, of massive pulmonary hemorrhage. The pneumothorax was of only thirty-five days' duration and was done for the control of pulmonary bleeding; the other one died of tuberculosis thirty days after delivery. The pneumothorax was of only five days' duration, and was done for the control of pulmonary hemorrhage. Both of these patients probably would have died of hemorrhage during labor if they had not had cesarean section. The third one had controlled pulmonary disease with negative sputum, but active pleurisy with effusion (positive for tubercle bacilli) complicating the pneumothorax. She has been lost to follow-up study, but the Tennessee Bureau of Vital Statistics has no record of her death after three years.

There were 2 patients whose lungs were expanded after artificial pneumothorax, and one of these did satisfactorily after delivery and one was worse. The one that was worse had the lung re-expanded because the collapse was unsatisfactory, and the opposite lung had far-advanced disease. She was extremely sick at the time of delivery and is presumed to be dead. There was no follow-up on her.

There were 3 patients in Group 1 who had had thoracoplasties and all 3 of them had satisfactory postoperative courses.

There were 4 patients in Group 1 who had phrenic paralysis; 1 of them had a satisfactory postoperative course, and the other 3 were worse. Two of the patients whose postoperative courses were worse had bilateral far-advanced disease with positive sputum and hopeless prognosis. One of them died of tuberculous meningitis three months after operation. The other died of tuberculosis nine months after operation. The third one had moderately advanced disease, and died of tuberculosis twenty-seven months after operation.

TABLE II. GROUP 2

CESAREAN SECTION WITHOUT COLLAPSE							
	NO. OF CASES	POST-OPERATIVE COURSE SATISFAC-TORY	POST-OPERATIVE COURSE WORSE	MATERNAL DEATHS FROM DELIVERY	DIED LATER	PROB-ABLY DIED LATER	ALIVE 11 YEARS LATER QUIESCENT
Totals	21	19	2	0	1*	0	1†

*Died of tuberculosis three months after operation. She had an acute exudative extension of her disease.

†Alive eleven years later with a positive sputum. She is doing part of her housework.

There were 21 cases in Group 2 (Table II); 19 of these patients had satisfactory postoperative courses, and 2 were worse. Both of those that were worse had active bilateral disease with positive sputum; one of them had acute exudative extension of the disease and died of tuberculosis three months after operation. The other is alive eleven years after delivery but still has a positive sputum although she is doing part of her housework.

There were 15 patients in Group 3 (Table III); all of them had satisfactory post-partum courses.

TABLE III. GROUP 3

FORCEPS DELIVERY WITH COLLAPSE						
	NO. OF CASES	POST-PARTUM COURSE SATISFACTORY	POST-PARTUM COURSE WORSE	MATERNAL DEATHS FROM DELIVERY	DIED LATER	PROBABLY DIED LATER
Art. Pneumo. without continuous caudal	5	5	0	0	0	0
Art. Pneumo. with continuous caudal or spinal	7	7	0	0	0	0
Thoracoplasty	2	2	0	0	0	0
Phrenic	1	1	0	0	0	0
Totals	15	15	0	0	0	0

Group 4 (Table IV) consisted of 12 patients, 10 of whom had satisfactory post-partum courses; 2 of them were worse. One of these two had bilateral far-advanced active exudative disease with positive sputum and she died of tuberculosis seven months later. The disease was hematogenous and resembled miliary tuberculosis. The other had moderately advanced exudative disease with positive sputum and congestive heart failure. There is no record of her death in the Tennessee Bureau of Vital Statistics, and we have lost track of her. We presume she is dead. Four of this group had continuous caudal or spinal analgesia, and all 4 had satisfactory post-partum courses. There were 11 forceps deliveries in Groups 3 and 4, in which forceps were employed merely for the final extraction of the baby after dilatation was complete. These patients had continuous caudal or spinal analgesia and were unable to expel the baby because of the anesthesia.

TABLE IV. GROUP 4

FORCEPS DELIVERY WITHOUT COLLAPSE						
	NO. OF CASES	POST-PARTUM COURSE SATISFACTORY	POST-PARTUM COURSE WORSE	MATERNAL DEATHS FROM DELIVERY	DIED LATER	PROBABLY DIED LATER
Without continuous caudal	8	6	2	0	1*	1†
With continuous caudal or spinal	4	4	0	0	0	0
Totals	12	10	2	0	1	1

*Died of tuberculosis seven months after delivery. At the time of delivery the disease was hematogenous and acute.

†Had moderately advanced active exudative disease with congestive heart failure at the time of delivery and is presumed to be dead although no record of her death can be found.

Group 5 (Table V) consisted of 9 patients, 8 of whom had satisfactory post-partum courses. The 1 patient whose post-partum course was worse was one of 7 pneumothorax cases. She had far-advanced right lower lobe disease with a large unclosed cavity and positive sputum. She died of tuberculosis twelve months later. There was 1 patient each in this group who had a

TABLE V. GROUP 5

SPONTANEOUS DELIVERY WITH COLLAPSE						
	NO. OF CASES	POST-PARTUM COURSE SATISFACTORY	POST-PARTUM COURSE WORSE	MATERNAL DEATHS FROM DELIVERY	DIED LATER	PROBABLY DIED LATER
Artificial pneumothorax	7	6	1	0	1*	0
Thoracoplasty	1	1	0	0	0	0
Phrenic	1	1	0	0	0	0
Totals	9	8	1	0	1	0

*She died of tuberculosis twelve months later. She had an unclosed cavity in the right base and positive sputum.

thoracoplasty and a temporary phrenic paralysis; both of them had satisfactory post-partum courses.

Group 6 (Table VI) consisted of 20 patients, 18 of whom had satisfactory post-partum courses. The 2 whose post-partum courses were worse had far-advanced bilateral active tuberculosis with positive sputum. One of them had far-advanced bilateral exudative disease with massive effusion on the left, and

TABLE VI. GROUP 6

SPONTANEOUS DELIVERY WITHOUT COLLAPSE						
	NO. OF CASES	POST-PARTUM COURSE SATISFACTORY	POST-PARTUM COURSE WORSE	MATERNAL DEATHS FROM DELIVERY	DIED LATER	PROBABLY DIED LATER
Totals	20	18	2	0	2*	0

*One of these had bilateral far-advanced exudative disease and massive effusion on the left. She died six months later of tuberculous meningitis. The other had far-advanced bilateral proliferative disease with cavities in both lungs. She died of tuberculosis thirty-nine days after delivery. Each of these patients had a hopeless prognosis.

the other had far-advanced bilateral proliferative disease with multiple cavities. Each was considered to have a hopeless prognosis.

Group 7 (Table VII) consisted of 4 patients, and there was 1 each who had artificial pneumothorax, thoracoplasty, phrenic paralysis, and no collapse.

TABLE VII. GROUP 7

THERAPEUTIC INTERRUPTION OF PREGNANCY, HYSTEROTOMY							
	NO. OF CASES	POST-OPERATIVE COURSE SATISFACTORY	POST-OPERATIVE COURSE WORSE	MATERNAL DEATHS FROM DELIVERY	NOT ACCOUNTED FOR	DIED LATER	PROBABLY DIED LATER
Artificial pneumothorax	1	1*	0	0	0	1	0
Thoracoplasty	1	1†	0	0	0	0	0
Phrenic	1	1‡	0	0	0	0	0
No collapse	1	1§	0	0	0	0	0
Totals	4	4	0	0	0	1	0

*Died seven and one-half years later of tuberculosis.
†Progress satisfactory nine months later.
‡Disease has spread and she is worse three years later.
§Disease arrested ten years after hysterotomy.

All of them had hysterotomies, and they all did satisfactorily after their operations. Therapeutic interruption was considered indicated because in one case there was active exudative moderately advanced pulmonary tuberculosis, with positive sputum, rheumatic heart disease, and syphilis; another had far-advanced bilateral disease with a thoracoplasty on the left and the disease was so widespread that we feared she could not undergo any of the methods of delivery without great danger to herself. We were influenced greatly in the latter case by the fact that her vital capacity was only about 1,200 c.c. and she occasionally had slight elevations of temperature. She had a hysterotomy and sterilization. The other two had active bilateral far-advanced proliferative disease with multiple cavities and a considerable amount of fever daily. We felt that they probably would not survive the period of their pregnancy and that they would probably abort spontaneously because of the high fever. They had grown worse after becoming pregnant, which was further reason for emptying the uterus. The patient (see footnote to Table VII‡) in this group who had a temporary phrenic paralysis with the exudative pulmonary tuberculosis, rheumatic heart disease, and syphilis is alive three years later, but the tuberculosis has spread considerably and her outlook is poor. The one (see footnote to Table VII†) who had had a thoracoplasty and bilateral far-advanced disease is progressing satisfactorily nine months after operation. One of those (see footnote to Table VII) with far-advanced bilateral proliferative disease with pneumothorax and high daily fever died of tuberculosis seven and one-half years later. The one with no collapse (see footnote to Table VII§) is reported to have the disease arrested about ten years later.

Group 8 (Table VIII) consisted of three patients. Two of them had syphilis which was considered the cause of the spontaneous abortion and no cause was discoverable for the other one. They have not been followed.

TABLE VIII. GROUP 8

	SPONTANEOUS ABORTION			
	NO. OF CASES	POST-PARTUM COURSE SATISFACTORY	POST-PARTUM COURSE WORSE	MATERNAL DEATHS FROM DELIVERY
Artificial pneumothorax	1	1	0	0
No collapse	2	2	0	0
Totals	3	3	0	0

No follow-up on any of this group.

COMPLICATIONS

Eight of our tuberculous patients had other complications than pregnancy.

1. One (Case 77) had a pleural effusion containing tubercle bacilli before cesarean section, complicating an artificial pneumothorax of nine months' duration. She had proliferative pulmonary disease with negative sputum.

2. One (Case 90) had a nontuberculous wound infection following cesarean section. She also had pleural effusion. The pulmonary disease was proliferative and it was eventually controlled as was the pleural effusion.

3. One (Case 6) developed tuberculous empyema nine months after cesarean section. The artificial pneumothorax was of only one month's duration at the

time of delivery. The pulmonary disease was proliferative, far-advanced unilateral.

4. One (Case 52) had reactivation of arrested tuberculosis with spread to the opposite side. The baby was delivered by forceps without continuous caudal or spinal analgesia.

5. One (Case 58) had tuberculous peritonitis and tuberculous pleurisy complicating the pneumothorax at the time of the cesarean section.

6. One (Case 64) had massive collapse of the diseased lung following cesarean section. She had no therapeutic collapse and the postoperative course otherwise was not remarkable.

7. One (Case 94) had tuberculous meningitis two months after delivery by cesarean section. She had had phrenic paralysis for two months at the time of delivery. The disease was proliferative and far advanced.

8. One (Case 3) had tuberculosis of spine and kidney and congestive heart failure. She was delivered by low forceps; no follow-up was made. The disease was far-advanced proliferative.

DISCUSSION

It is worthy of note that only 4 of our 118 patients had the pregnancies interrupted deliberately.

It is clear that the 14 patients who were worse after delivery were practically hopelessly sick with tuberculosis and had little, if any, chance for survival even if they had not been pregnant. The fact that two of them are alive is testimony to the efficacy of the method by which they were managed.

It will be seen, by comparing tables, that of the 14 whose progress was worse after delivery, there were 7 of 34 who had cesarean section with collapse (20.6 per cent); and that 2 of 21 had cesarean section without collapse (9.4 per cent); none of the 14 had forceps delivery with collapse; 2 of 12 had forceps delivery without collapse (16.6 per cent), and both of them were delivered without continuous caudal or spinal analgesia; 1 of 9 had spontaneous delivery with collapse (11.1 per cent); 2 of 20 had spontaneous delivery without collapse (10 per cent). One might conclude from these findings that cesarean section delivery was the least satisfactory method and that spontaneous delivery was the best. While this might seem true, it is not so if we consider the fact that the patients in the cesarean section delivery group had the poorest prognosis from the standpoint of the tuberculosis, and that those in the spontaneous group had the best prognosis. The entire forceps delivery group of patients had almost the same percentage of unfavorable courses after delivery as the spontaneous delivery group, but when we separate the continuous caudal or spinal patients from those without continuous analgesia, there is a marked difference; none of the patients in the continuous caudal and spinal group was worse, while the rest of the forceps group had 12.5 per cent worse. It is noteworthy that the prognosis of patients in this entire group occupied an intermediate position with the other two groups; that is, the cesarean delivery group and the spontaneous delivery group. In fact, the progress of the three groups reflects the prognosis from the standpoint of the tuberculosis.

The 97 patients in our series who did satisfactorily after delivery were classified when they came under our observation according to the N.T.A. Classification as follows: 23 were far-advanced active with mainly proliferative disease and 16 were far-advanced active with mainly exudative disease; 17 were moderately advanced active with mainly proliferative disease, and 2 were moderately advanced active with mainly exudative disease; 4 were minimal active with mainly proliferative disease; 11 were far-advanced inactive with mainly proliferative disease; 22 were moderately advanced inactive with mainly proliferative disease; and 12 were minimal inactive with mainly proliferative disease. All those patients who could not be classified as having disease arrested were classified as active because we felt that if they had not been stable long enough to be considered arrested, the effect of labor would likely be the same as it would if they were "frankly" active.

Seventeen of the 97 patients who did satisfactorily after delivery had positive sputum at the time their babies were born (Table IX). Three of them had exudative disease which agrees with the experience of Ornstein and Kovnat[125, 126] and others that those with exudative tuberculosis may stand pregnancy well. Fourteen of these patients had mainly productive disease with some exudate. Eleven had satisfactory *artificial pneumothorax* and 1 of these had exudative disease. Of these 11, 5 were delivered by cesarean section, 4 by forceps, and 2 spontaneously. Two of these positive sputum patients had *thoracoplasty* and both had productive lesions; 1 was delivered by cesarean section and 1 delivered spontaneously. One of this group had *phrenic paralysis* and delivered spontaneously. There were 3 patients in this positive sputum group who had *no collapse*, but were classified as quiescent. Two of them had exudative disease and 1 had the productive type; 1 of these was delivered by cesarean section and 2 by forceps.

The difference in the post-partum course of this latter group and the group of 14 patients who did poorly after delivery seems to be that they had the disease controlled in contrast to the fact that the other group had progressive uncontrolled disease. And furthermore, we believe that the way in which these 17 cases were managed contributed a lot to their favorable progress.

TABLE IX

CLASSIFICATION OF NINETY-SEVEN PATIENTS WITH SATISFACTORY POST-PARTUM COURSES		
Far Advanced:		
Active, mainly proliferative	23	
Active, mainly exudative	6	
Inactive, mainly proliferative	11	
Total		40
Moderately Advanced:		
Active, mainly proliferative	17	
Active, mainly exudative	2	
Inactive mainly proliferative	22	
Total		41
Minimal:		
Active, mainly proliferative	4	
Inactive, mainly proliferative	12	
Total		16

In our series of 118 cases, we experienced a mortality rate of 9.3 per cent, which appears to be extremely low when compared to a series of 85 cases studied by Ornstein Ulmar and Dittler as reported by Falls, which experienced a mortality rate of 36 per cent in five years, and is favorably comparable to a series of 82 cases reported by Ornstein and Epstein[128] which experienced a mortality rate of 14.6 per cent. If we count as dead the two patients who probably died, our mortality rate would be 11 per cent.

We are inclined to consider these statistics of little significance. It is our opinion that none of these patients had the course of the tuberculosis influenced adversely by the methods of delivery employed since those who died and those who are presumed to have died would certainly have done so even if they had not been pregnant. The extent of the disease and the degree of the activity would have made such an outcome almost inevitable.

SUMMARY

We believe from our experience that the methods of delivery we have employed are satisfactory. Despite the fact that our patients with cesarean section delivery had the worst record, we believe this method is the one of choice in the worst cases. Objection has been voiced to this procedure because of the high mortality rate from cesarean section. The answer to this objection is that the mortality rates from this procedure are based on all types of cesarean deliveries, including emergencies, those patients who have been in labor many days, those who have had presenting hands, cords, feets, etc., and those who have had one or more vaginal examinations.

We decide early in the course of pregnancy with our patients that cesarean section will be the method of delivery, and their care is directed toward this end. They are delivered at term or just before term, and are sterilized if further pregnancies are considered undesirable or dangerous. The fact that we had no operative mortality in 55 patients is testimony to the safety of this procedure when employed deliberately in patients who are properly prepared for it. A close second, in our opinion, is delivery by continuous caudal or continuous spinal analgesia with low forceps extraction, when dilatation is complete. It is possible that further experience may lead us to believe that this method is better than cesarean section. When the anesthesia is satisfactory, the patients experience no inconvenience or pain and are not exhausted by the delivery.

While our tables show good results in those patients delivered by forceps without continuous caudal or continuous spinal analgesia, we believe it is not a good method of delivery in active and uncontrolled tuberculosis, because there is some bearing down and consequent increased intrapulmonary pressure during the first stage of labor.

We are convinced, from our experience, that therapeutic interruptions of pregnancy are seldom justified because of pulmonary tuberculosis and should be reserved for those patients in whom pregnancy itself is difficult and those in whom the other complications of pregnancy are serious. It should be used

in those cases of pulmonary tuberculosis in which the patient shows little resistance to the tuberculosis and is obviously or apparently unable to cope with the disease. It should not be employed after the third month of pregnancy because it is our observation and that of others[18, 34, 140, 153] that after the first trimester therapeutic interruption of pregnancy is as serious as delivery at full term and probably more so than delivery by one of the methods we have discussed here.

While we are impressed by the observation that[125, 126, 128] only exudative and caseous pneumonic cases do badly with pregnancy, we note that three of our patients who did well after delivery had exudative tuberculosis and positive sputum. We believe, however, that they would not have had satisfactory progress if they had not been controlled as outlined here; we also note that most of our patients who were worse after delivery had proliferative disease that was progressive; therefore, we believe that control of the disease is of more importance than the type of the disease.

We have tried to develop indications for the different methods of delivery and we believe the patients with the worse prognosis should have cesarean section. We believe further that continuous caudal analgesia and continuous spinal analgesia should be used in patients with better prognosis where there is less danger of reactivation of the tuberculosis. This would include those whose tuberculosis is quiescent by the N.T.A. Classification. Forceps delivery without continuous caudal or spinal analgesia may be used in quiescent or better cases where the analgesia is not effective and it is desirable to avoid the second stage of labor. Spontaneous delivery should be reserved for multiparous women whose tuberculosis has been arrested for more than one year, and whose histories indicate short easy labors. Primipara may be allowed to deliver spontaneously if the disease is apparently cured. Therapeutic interruption of pregnancy should never be done except in extreme cases in which pregnancy without tuberculosis would be difficult, and in those in whom the other complications are serious.

It is most difficult to show that the second stage of labor, with its increased intrapulmonary pressure, is responsible for the harmful effects of pregnancy and the puerperium; however, the fact that 52 of our 97 patients with satisfactory post-partum courses were active and 17 of them had positive sputum lends support to the belief when we remember that their deliveries were conducted in such a way as to eliminate or minimize the effects of this pressure.

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SPONTANEOUS COLLAPSE

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THE infiltration of air into tissue and the bursting of air into the thoracic and abdominal cavities have been reported separately in medical literature. In this treatise the subject of the abnormal presence of air in different anatomic areas is examined connectively, since air often migrates or flows from one area to another. The presence of extraneous air has been diagnosed as interstitial emphysema, spontaneous mediastinal emphysema, subcutaneous emphysema, spontaneous pneumothorax, and in this report an exceptional condition is diagnosed as spontaneous pulmonary pneumoperitoneum. The presence of air and blood, or just blood, in the thoracic cavity has induced conditions which are characterized as hemorrhagic pleurisy, spontaneous hemopneumothorax, and spontaneous hemothorax. The latter diagnosis is made because air has been absorbed before its presence has been demonstrated. All of the aforementioned pneumatic conditions are not common or well known. The absence of essential air in pulmonary tissue occurs in spontaneous atelectatic (massive) collapse. This pathologic entity must be differentiated from spontaneous collapse caused by the presence of extraneous air.

SPONTANEOUS INTERSTITIAL EMPHYSEMA

Interstitial emphysema is the precursor of spontaneous mediastinal emphysema, spontaneous pneumothorax, and subcutaneous emphysema. When alveoli rupture, air spreads along the connective tissue bands, which surround the bronchi and blood vessels of the lung. This may occur without trauma or markedly increased intrapulmonary pressure. It does occur after trauma, after some postoperative procedures, and with pulmonary infections. It is often associated with bronchopneumonia, influenza, pertussis, measles, and bronchial asthma. Congenitally thin alveolar walls, or walls weakened by disease, collapse spontaneously when intrapulmonary pressure is increased by coughing or straining. Uncontrollable cough caused the rupture of alveoli in the cases herein reported. On the other hand, interstitial emphysema may be one of the causes of incessant coughing. Macklin[1] demonstrated interstitial emphysema, which is the genesis of spontaneous mediastinal emphysema, by artificially increasing the intrapulmonary pressure in animals. He showed that air enters the perivascular sheaths of the finer branches of the pulmonary vessels. As the intrapulmonary pressure increases, the bubbles of air coalesce into larger ones and move toward the root of the lung. At the root, the air bubbles merge into large blebs which rupture into the mediastinum or produce a pneumothorax. Extraneous air may do both simultaneously. Ballon and Francis[2] and Macklin[1] described the course of air into the neck, face, axilla, anterior chest wall, arms, alongside of the

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trachea, elevating the tongue, or downward into the retroperitoneal space around the kidney. Fischer and Macklin[3] supported the theory that overdistention of the opposite lung could cause mediastinal emphysema.

The terms interstitial and mediastinal emphysema have been employed interchangeably in the literature. Reports on cases of subcutaneous emphysema and surgical emphysema discuss interstitial and mediastinal emphysema. Interstitial emphysema per se cannot be described clinically. Subcutaneous emphysema is often a definite sign of interstitial and mediastinal emphysema. Therefore, the general use of the term mediastinal emphysema, which can be demonstrated by both physical and x-ray examination, would at least clarify topic nomenclature.

SPONTANEOUS MEDIASTINAL EMPHYSEMA

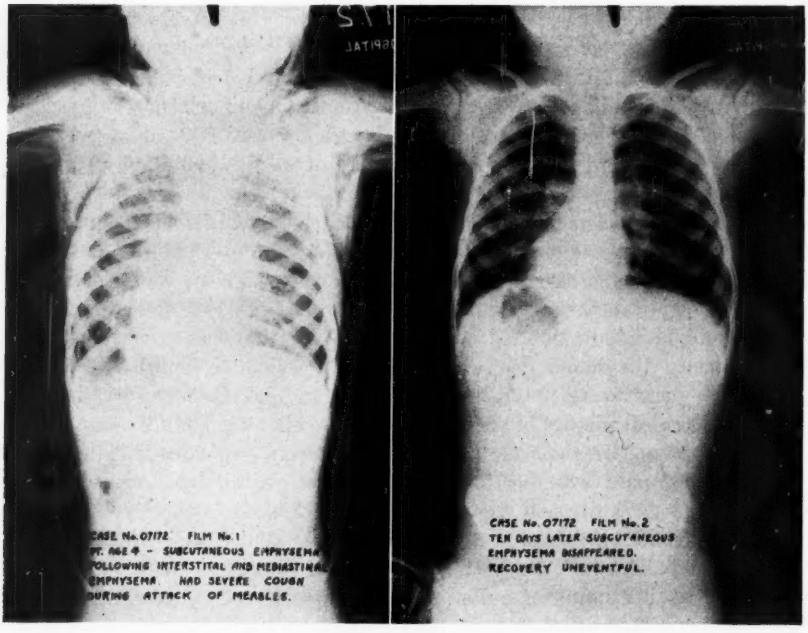
In 1831 Laennec[4] described curious grating sounds and bubbling râles during respiration as diagnostic signs of subpleural and interlobar emphysema of the lung. These signs were confirmed by Skoda[5] in 1850. Six years later Rokitansky[6] described the pathologic anatomy of mediastinal emphysema which followed pulmonary trauma and ulceration of the trachea.

Muller[7] in 1888 described physical signs characteristic of pneumomediastinum: namely, the presence over the precordium of bubbling crepitations synchronous with the heartbeat. He said that the diagnosis was greatly facilitated by the appearance of subcutaneous emphysema. In 1918 Rees and Hughes,[8] Smith,[9] and Munden[10] pointed out the diagnostic significance of these sounds. Smith termed the sound "pericardial knock" which he heard over the precordium in penetrating wounds of the chest. In 1937 Hamman[11] called attention to the clinical picture of six cases which had spontaneous air occurring over the mediastinum. In the same year Scott[12] reported two cases and thought the accompanying pain was due to angina pectoris. Additional cases have been recognized by McGuire and Bean,[13] Morey and Sosman,[14] Wolfe,[15] Matthews,[16] Pinckney,[17] Caldwell,[18] Styron,[19] Miller,[20] Murphy and Zeis,[21] Griffin,[22] Lintz,[23] Greene,[24] and Adcock.[25] In 1944 Miller[26] added four more cases, two of which demonstrated this condition and an associated spontaneous pneumothorax.

Graham, Singer, and Ballon[27] observed mediastinal emphysema after pulmonary lobectomy. Sercer and Peicic[28] said it occurred during pneumothorax. Parfitt and Crombie[29] reported the same condition when the lung was injured. Alexander[30] reported a case of tuberculosis which developed interstitial, mediastinal, and subcutaneous emphysema after spontaneous collapse of the involved lung. Pneumothorax had been instituted, but the collapse was not due to trauma. Jehn and Nissen,[31] and Banyai and Jurgens[32] have reported mediastinal emphysema following pneumoperitoneum. Since the emphysema occurred following injection of air by the subphrenic route and did not occur when air was given by the subumbilical route, the pneumomediastinum was due to misplaced air dissecting upward under the basal parietal pleura.

Graebner[33] reported three cases of pneumomediastinum and two of pneumopericardium. In one case of pneumopericardium tracheotomy had been performed twenty-four hours previously. In the other case pneumopericardium

occurred without tracheotomy. Both had acute obstructive laryngitis. He did not know how air entered the pericardial sac and reported that Macklin had never found this condition in his experiments. It is doubtful whether mediastinal air with its origin from interstitial emphysema can extend into the pericardial space for anatomic reasons. Beck[34] said that an opening in the pericardium would have to be present to admit air. Etiologic factors of pneumopericardium are trauma, pericarditis, infection with gas bacillus, erosion and perforation of disease processes from neighboring air-containing organs, radium necrosis, and congenital defects.


CASE No. 07172 FILM No. 1
PT. AGE 9 - SUBCUTANEOUS EMPHYSEMA
FOLLOWING INTERSTITAL AND MEDIASTINAL
EMPHYSEMA. HAD SEVERE COUGH
DURING ATTACK OF MEASLES.
CASE No. 07172 FILM No. 2
TEN DAYS LATER SUBCUTANEOUS
EMPHYSEMA DISAPPEARED.
RECOVERY UNEVENTFUL.

A. *B.*

Fig. 1.—*A*, Subcutaneous emphysema following interstitial and mediastinal emphysema. *B*, Ten days after subcutaneous emphysema had disappeared.

Clinically, mediastinal emphysema may be suspected by palpating the subcutaneous tissue of the neck. This extraneous air gives the patient a tight feeling about the neck, even when it is confined to one side. Of course, the subjective feeling of compression is much more distressing when air infiltrates the neck bilaterally. The patient is apprehensive and complains of choking and difficulty in deglutition. Pain and distress referred to the mediastinum are commensurate with the amount of distention and pressure. Macklin[1] thought that, when the initial pain is lateral, the origin is in the interstitial tissue or pleura rather than in the mediastinum. The migration of air into the neck and subcutaneous tissue acts as a safety valve, and prevents tamponade on the

heart and subsequent circulatory embarrassment. In animal experiments Ballon and Francis[2] noted that when mediastinal emphysema was marked, subcutaneous emphysema was slight, and vice versa. Most clinicians point out that the pathognomonic sign of mediastinal emphysema is the crunching, crackling, popping sign which is synchronous with the heartbeat.

In this report two cases are presented. Case 1 (Fig. 1, *A*), a boy, aged 4 years, was admitted without x-ray evidence of intrathoracic air, but with a marked subcutaneous emphysema. Fig. 1, *B* shows an x-ray view of the same patient ten days after symptoms had subsided; no air was present. In the second case, a boy 13 years of age (Fig. 2, *A*), the radiograph shows a bilateral bronchitis and bronchiolitis. A small area of atelectasis is present in the right lower lobe. Fig. 2, *B* shows a radiograph immediately after bronchoscopy. The segmental atelectasis in the lower lobe had disappeared. Fig. 2, *C* shows a radiograph taken twenty-four hours after bronchoscopy. Thirty minutes after bronchoscopy subcutaneous emphysema was evident. A left spontaneous pneumothorax remained twenty-four days after the occurrence of mediastinal emphysema. The patient made an uneventful recovery and the uncontrollable cough disappeared.

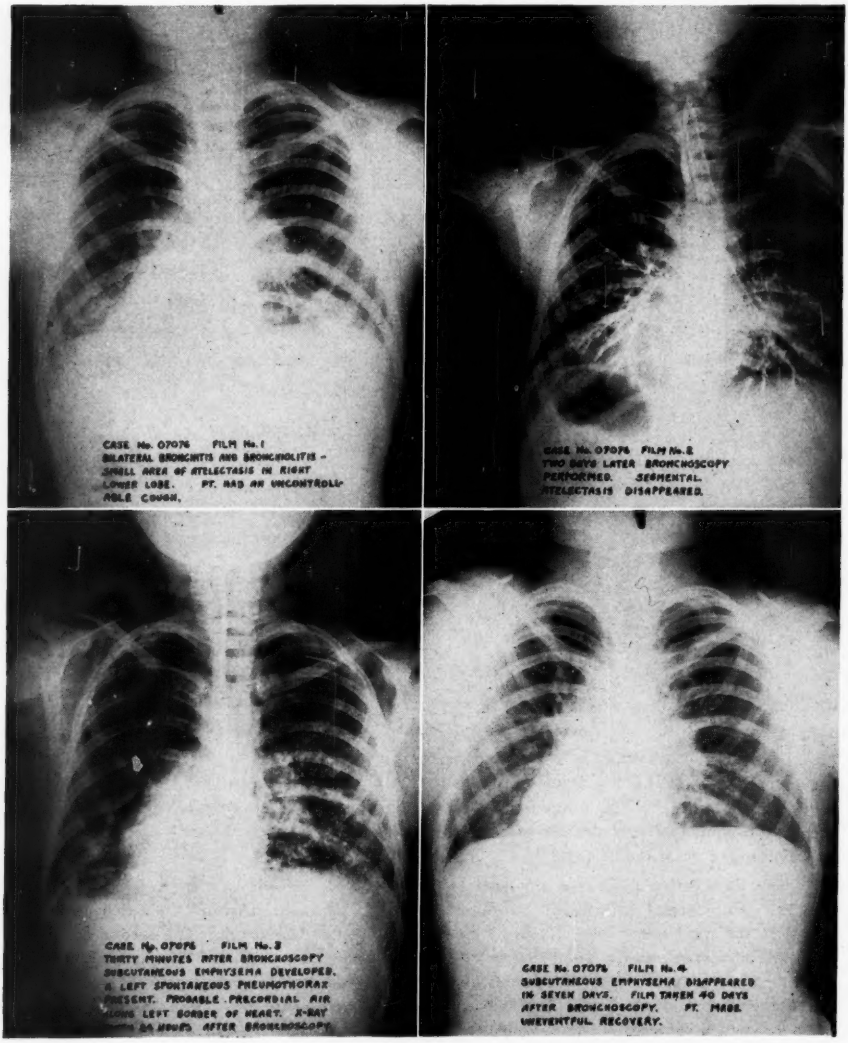
CASE 1 (Fig. 1, *A* and *B*).—The patient, a boy aged 4 years, was admitted with a generalized subcutaneous emphysema of twenty-four hours' duration. He complained of tightness around the neck and some shortness of breath. Temperature was 100.2° F., pulse 120. X-ray picture demonstrated subcutaneous emphysema, but no spontaneous pneumothorax. Heart sounds were not accompanied by any crunching sounds, since the air had disappeared into the subcutaneous tissues, as evidenced by x-ray. The cause of the initial interstitial and mediastinal emphysema was due to excessive coughing while the patient was convalescing from an attack of measles. After five days of hospitalization, the patient was comfortable and made an uneventful recovery.

CASE 2 (Fig. 2, *A*, *B*, *C*, and *D*).—The patient, aged 13 years, white, male, overweight, was admitted to the hospital with a history of a pulmonary illness of seven weeks' duration. Temperature was 102° F., pulse 110, respiration 23. He had violent coughing attacks and was raising a moderate quantity of sputum. The sputum was negative for any specific organism, but a few gram-negative, rod-shaped diplococci were observed. The white blood cell count was 11,500. A diagnosis of bilateral bronchiolitis was made. After the bronchoscopy a course of sulfonamide and penicillin was administered. In two weeks the patient was discharged from the hospital.

Ten days after discharge the patient re-entered the hospital, with a temperature of 101° F. Paroxysmal attacks of coughing persisted. A small area of atelectasis was observed in the right lower lobe. It was impossible to control the cough. The second bronchoscopy was unventful, and a bronchogram showed the atelectatic area had disappeared. Thirty minutes after bronchoscopy the patient experienced dyspnea. He complained of choking and of not being able to breathe. The subcutaneous tissues of the chest, neck, and entire face were distended with air. The eyelids puffed out and interfered with vision. A crunching sound was audible over the mediastinum. He was placed in an oxygen tent. A Levine tube was inserted into the esophagus for feeding, since the patient claimed he could not swallow. The pulse increased to 120, and temperature averaged 101° F. for three days. The subjective symptoms subsided in four days. Subcutaneous air disappeared in seven days. Recovery was uneventful. Bronchoscopy probably hastened the occurrence of the mediastinal and sub-

cutaneous emphysema. Interstitial emphysema was present, no doubt, prior to the bronchoscopy and may have been responsible for the most uncontrollable cough ever witnessed in a patient by the author.

A. *B.*


CASE No. 07076 FILM No. 1
BILATERAL BRONCHITIS AND BRONCHIOLITIS -
SMALL AREA OF ATELECTASIS IN RIGHT
LOWER LOBE. PT. HAD AN UNCONTROLL-
ABLE COUGH.
CASE No. 07076 FILM No. 2
TWO DAYS LATER BRONCHOSCOPY
PERFORMED. SEGMENTAL
ATELECTASIS DISAPPEARED.
CASE No. 07076 FILM No. 3
THIRTY MINUTES AFTER BRONCHOSCOPY
SUBCUTANEOUS EMPHYSEMA DEVELOPED.
A LEFT SPONTANEOUS PNEUMOTHORAX
PRESENT. PROBABLE PRECORDIAL AIR
LEFT BORDER OF HEART. X-RAY
CASE No. 07076 FILM No. 4
SUBCUTANEOUS EMPHYSEMA DISAPPEARED
IN SEVEN DAYS. FILM TAKEN 40 DAYS
AFTER BRONCHOSCOPY. PT. MADE
UNEVENTFUL RECOVERY.

C. *D.*

Fig. 2.—*A*, bilateral bronchitis and bronchiolitis—segmental atelectasis in the right lower lobe. *B*, Bronchogram shows disappearance of area of segmental atelectasis. *C*, Twenty-four hours after bronchoscopy—subcutaneous emphysema and spontaneous pneumothorax on the left. *D*, Forty days after occurrence of interstitial mediastinal subcutaneous emphysema and spontaneous pneumothorax.

SPONTANEOUS PNEUMOTHORAX

Laennec[35] in 1819 discussed spontaneous pneumothorax, nontuberculous pneumothorax, and double pneumothorax, which is today diagnosed as bilateral simultaneous pneumothorax. In 1887 Hall[36] reported the first series of spontaneous pneumothorax in the healthy individual. Ri[illegible]nan and Fussel[37] (1902) and Emerson[38] (1903) were the first to report nontuberculous spontaneous pneumothorax in this country. At this time it was thought the etiologic factor was tuberculosis. Our present concept concedes that the most common etiologic factors are emphysematous blebs in the pleura. Gordon[39] described five cases in which, after re-expansion, he could delineate the bullae. Kirschner[40] reported twenty cases of spontaneous pneumothorax, and in one patient, who died of acute cardiac failure, he found a rupture of a pleural bleb.

In 1931 Kjaergaard[41] wrote extensively on spontaneous pneumothorax in the apparently healthy. In the same year McMahon[42] reported a case of spontaneous bilateral pneumothorax. Glickman and Schlomovitz[43] reported a case of simultaneous bilateral pneumothorax complicating pneumoconiosis, which brought the number of bilateral simultaneous pneumothorax cases to eighty-two. Gaetan[44] reported a case of simultaneous bilateral pneumothorax due to tuberculosis in 1942. Superimposed spontaneous pneumothorax occurs more frequently than is generally believed, since it happens in many patients receiving therapeutic treatment for tuberculosis. McCloskey[45] reported on a series of 1,131 cases of induced pneumothorax. These cases had 52,835 refills over a period of six years, and this condition was noted 161 times, or 14.2 per cent.

Charr[46] cited eleven cases of spontaneous pneumothorax; Schomer and Ehrlick[47] (1935) six cases; Ehrlick and Schomer[48] (1938) seven cases; Hedgpeth and co-workers[49] (1944) twenty cases; Babington[50] (1944) forty-four cases, including thirteen of his own; Hadley[51] (1941) five cases. Griffin[52] (1942) wrote that the incidence of spontaneous idiopathic pneumothorax is one case per thousand in male students. Englehardt and Derbes[53] reported two cases of spontaneous pneumothorax caused by bronchial asthma. The thinning of the alveoli wall was verified at autopsy.

The average case of idiopathic spontaneous pneumothorax demands few therapeutic suggestions, but spontaneous ruptures caused by pulmonary disease require urgent and immediate treatment. Tuberculosis is the etiologic factor involved in the majority of cases of pathologic spontaneous pneumothorax. Chronic emphysema, interstitial and mediastinal emphysema, pneumoconiosis, bronchopneumonia, bronchitis, asthma, congenital cysts, and tumors are other etiologic factors.

Mild cases of idiopathic spontaneous pneumothorax are diagnosed pleurisy, myositis, or intercostal neuralgia, especially if there is no recurrence. Oechsli and Skillen[54] reported a case of spontaneous pneumothorax with predominantly abdominal symptoms of pain, nausea, and vomiting due to downward retraction of the diaphragm. Joress[55] reported two cases with similar symptoms.

Clinically, the onset in pulmonary collapse is usually sudden. The course of idiopathic spontaneous pneumothorax is benign and afebrile, often with a transudate, but not exudate. Pain and shortness of breath make the patient realize that something has happened. Pain is exaggerated during inspiration and may increase as the intrathoracic pressure increases. If the valvelike opening in the pleura closes, the pressure may become neutralized and symptoms may ultimately disappear, even without re-expansion of the lung. The duration of a tension pneumothorax is proportional to the length of time it takes the opening in the visceral pleura to close. In this series, one patient had a pneumothorax for eight months after the tension pneumothorax had run its brief course. The continuation of the pneumothorax was probably due to leakage of air into the thoracic cavity intermittently through a valve-like opening in the lung. This is the cause of a prolonged pneumothorax.

Several authors [49,50,56,57] report a few cases in which lungs have remained collapsed for as long as seven and one-half years and ultimately have re-expanded. In other cases, re-expansion has failed to occur and the patients have had varying degrees of disability for years. Bassel[58] has reported the case of a patient who probably had an unexpanded lung for seventeen years, and who underwent a successful major surgical procedure (gastroenterostomy).

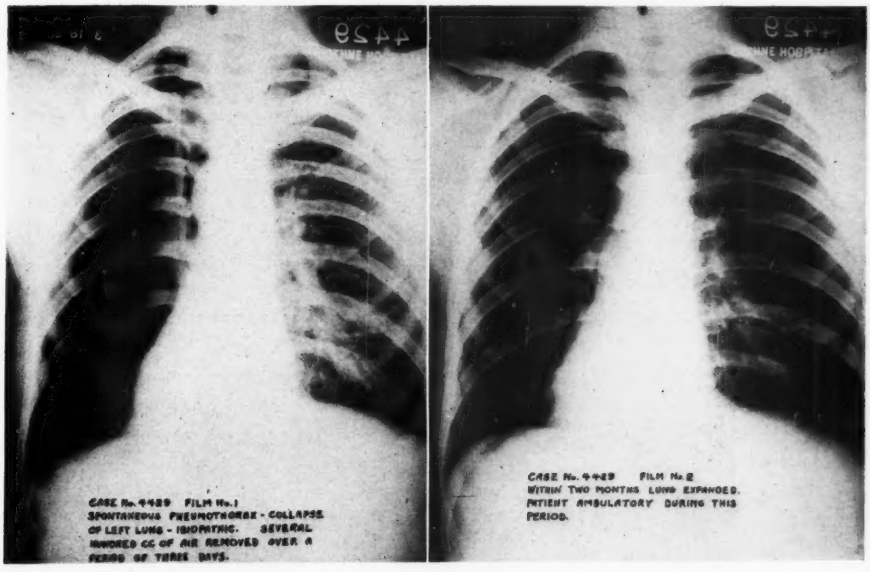
The course of pathologic spontaneous pneumothorax is usually severe. In those patients who have pathologic spontaneous pneumothorax, cyanosis and shock often occur, especially if there is a mediastinal shift, pulmonary hemorrhage, or tension pneumothorax. Spontaneous pneumothorax due to trauma, such as fractured ribs, stab and gunshot wounds, or causes due to sudden pressure changes occur frequently. Hemothorax is often supplementary, and treatment for hemorrhage, as well as shock, is necessary. Holter and Horwitz[59] reported a case of spontaneous pneumothorax in an airplane. Wilcox and Foster-Carter[60] (1937) cited a case in which an air pilot died during the third attack.

In this study twelve cases are reported which had spontaneous pathologic pneumothorax. Four patients who had bilateral tuberculosis died. Of the remaining eight, five had pulmonary tuberculosis, and two bronchitis. One had silicosis, due to fluorspar dust, and later died. One patient with tuberculosis had a thoracoplasty and died with a spontaneous pneumothorax in the opposite lung one week after the operation.

Spontaneous idiopathic pneumothorax may occur in either sex at any age. Wilson[61] stated that it more frequently appears in male adults. In this series of twenty patients, twelve were male and eight were female. Fifteen occurred on the right side and five on the left side. None of these cases were bilateral, or alternating. In the twenty cases of idiopathic spontaneous pneumothorax, the average duration was three months, except one case which had eight recurrences over a period of five years. One patient had two occurrences in one month; one, four in one month; one, six in six months; and one, five in two months. Locke[62] reported one patient who had a recurrence of spontaneous pneumothorax eighteen times. Eleven patients had air removed at periodic

intervals for comfort, three were treated with an irritant oil,[63] and four received no treatment. All recovered. Fig. 3, *A* is a radiograph showing spontaneous collapse of idiopathic etiology. Fig. 3, *B* shows a radiograph of the same patient two months later. No treatment was indicated.

Tyson, Crandall, and Hanover[64] (1941) reported on the surgical treatment of one case of recurring idiopathic spontaneous pneumothorax due to a ruptured bleb. He placed a purse-string suture at the base of the cyst, drew it tightly, and removed the bleb.


CASE No. 4429 FILM No. 1
SPONTANEOUS PNEUMOTHORAX - COLLAPSE OF LEFT LUNG - IDIOPATHIC. SEVERAL HUNDRED CC OF AIR REMOVED OVER A PERIOD OF THREE DAYS.
CASE No. 4429 FILM No. 2
WITHIN TWO MONTHS LUNG EXPANDED. PATIENT AMBULATORY DURING THIS PERIOD.

A. *B.*

Fig. 3.—*A*, Radiograph shows idiopathic spontaneous pneumothorax. *B*, Re-expansion of lung two months after occurrence of idiopathic spontaneous pneumothorax.

Hoyer and Clagett[65] were the first to perform a lobectomy for chronic spontaneous pneumothorax. They removed the middle lobe of the right lung because of multiple emphysematous bullae.

Pulmonary re-expansion has been obtained in a number of cases by the introduction of irritant substances into the pleural cavity for the purpose of producing obliterative pleuritis (Kjaergaard,[66] Harvey,[67] Hennel and Steinberg,[68] and Morlock[69]).

Chandler[70] (1937) used Gomenol in olive oil to produce obliterative pleurisy. He used only 25 c.c. of 20 per cent Gomenol and olive oil to create an exudative reaction, which he thought aided in the re-expansion of the lung. He also used 50 per cent dextrose to create the same condition.

Without the knowledge of Chandler's work, I used peanut oil effectively. However, unlike Chandler, a larger quantity of an irritant and not a bland

CASE NO. 6252 FILM 1
IDIOPATHIC SPONTANEOUS PNEUMOTHORAX
LEFT. DURATION 8 MOS. DYSPNEA ON EXERTION.
CASE NO. 6252 - FILM 2
ASPIRATION OF AIR FAILED TO
RE-EXPAND LUNG. OLEOTHORAX INSTITUTED
CASE NO. 6252 FILM 3
FLUID ASPIRATED EVERY 2 WKS. FOR
3 MOS. THEN OIL AND FLUID MONTHLY FOR
5 MOS. AFTER 10 MOS. LUNG FAIRLY WELL
EXPANDED, WITH EXCEPTION OF SMALL
POCKET OF FLUID (2 OZ.) PT. COMFORTABLE
AND WORKING.

A. *B.* *C.*

Fig. 4.—*A*, Radiograph shows spontaneous, idiopathic pneumothorax of eight months' duration. *B*, Oleothorax instituted after aspiration of air failed to re-expand lung. *C*, Radiograph shows obliterative pleuritis which was initiated by oleothorax. Small pocket remains, which contained 2 ounces of fluid.

oil was used, endeavoring at all times to keep the entire thoracic space filled with oil or fluid. This insures a pleuritis over the entire lung surface. After the oil remains for two to four weeks, it is gradually withdrawn at weekly intervals, along with fluid, to aid in lung re-expansion. With this procedure and the obliterative pleuritis which is created by the irritant oil, the lung is re-expanded gradually. The pleuritis seals over the opening in the pleura. One case reported (Fig. 4, *A*) had a spontaneous pneumothorax of eight months' duration. Oleothorax was instituted (Fig. 4, *B*) and carried for eight months before considerable re-expansion of the lung occurred (Fig. 4, *C*). A pulmonary fistula which is too large for oleothorax must be treated by the constant removal of air with suction. In these cases, a suction pump[71] can be attached to a water faucet and connected by tube to a needle in the chest wall. Periodic or continuous suction is applied. One patient with tuberculosis had this treatment twenty-four hours a day for one week, at the end of which time the exudate sealed the visceral fistula. Thoracoplasty and a modified Schede operation were employed in closing the emphysema cavity.

SPONTANEOUS HEMOPNEUMOTHORAX

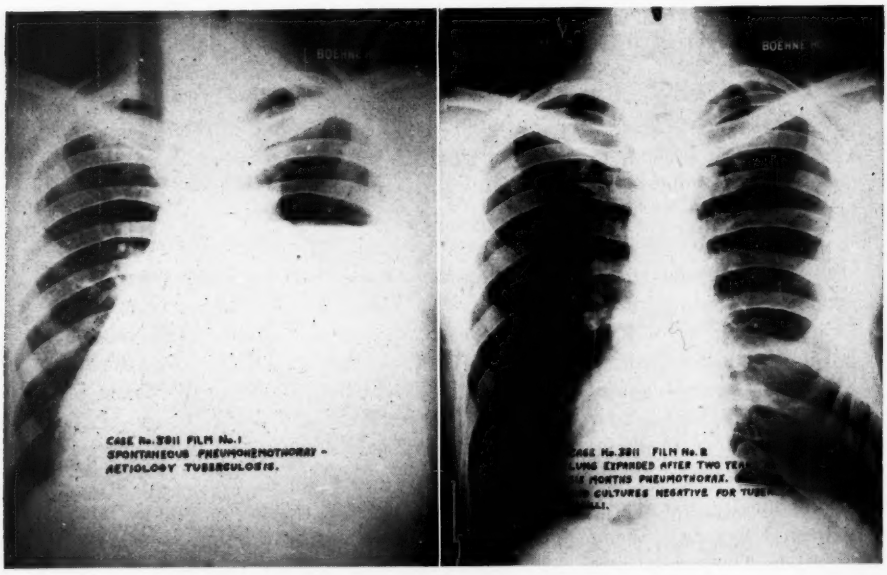
Nontraumatic spontaneous hemopneumothorax occurs in the same manner as spontaneous pneumothorax, except for the presence of hemorrhage. In fact, a spontaneous collapse by air probably precedes the hemorrhage. Often the air is absorbed when the hemothorax is discovered.

The quantity of blood in hemopneumothorax depends on the time it takes the collapsed lung to retract the bleeding vessel. Hemorrhage often occurs after the visceral pleura ruptures, and the bleeding vessel may not necessarily be in the opening of the pleura but in a torn vascular adhesion. Blood should not be left in the pleural cavity because the fibrin may later calcify. Irritation of the diaphragm caused by the blood and shifting of the mediastinum often bring about abdominal symptoms which require a differential diagnosis from appendicitis, perforated ulcer, or gall bladder disease. Not infrequently the kidney is investigated for the cause of the abdominal pain.

Clinically, the symptoms of hemopneumothorax consist of sudden onset of pain and dyspnea. Some improvement takes place until sign of excessive internal hemorrhage occurs. Abdominal symptoms may mask the lung collapse until extreme dyspnea suggests a roentgenogram of the chest. Physical signs of dullness and diminished breath sounds can be overlooked. The treatment consists of blood transfusion, with aspiration of the blood from the pleural cavity. Pneumothorax may be indicated. If compression by air does not stop the hemorrhage, an exploratory thoracotomy is advisable.

Hemopneumothorax not associated with trauma, heart disease, tumor, or pulmonary infarction is quite rare. In 1936 Jones and Gilbert[72] reviewed the literature on spontaneous hemopneumothorax and reported twenty-three cases from other clinical reports and one case of their own. Of the twenty-four cases, only one case had tuberculosis as a definite etiology. One patient reported in this series had tuberculosis. Tuberculosis was found in the hemorrhagic fluid

in cases reported by Coyon,[73] Hanot,[74] Montague,[75] Mainini,[76] Lancelin and Bideau,[77] Churnton,[78] Korol,[79] and Heise and Krause.[80] In the same year (1936) Korol[79] reviewed thirty-four cases of hemorrhagic pleurisy which occurred since 1900. Prior to 1900 verification by x-ray was not available. Korol[79] reported thirty cases of hemorrhage which occurred during artificial pneumothorax. In a series of 50,000 pneumothoraces, I have not seen a single case of hemorrhage in the pleural space which happened during the course of treatment. A hemorrhagic exudate seldom occurs if the lung is properly re-expanded. It does occur when an oleothorax is allowed to re-expand. Fluid which forms during re-expansion may become hemorrhagic. This may be due to the presence of granulation tissue, but more than likely it is due to the high negative pressure of some aspirations. It has been called *exudate ex vacuo.*



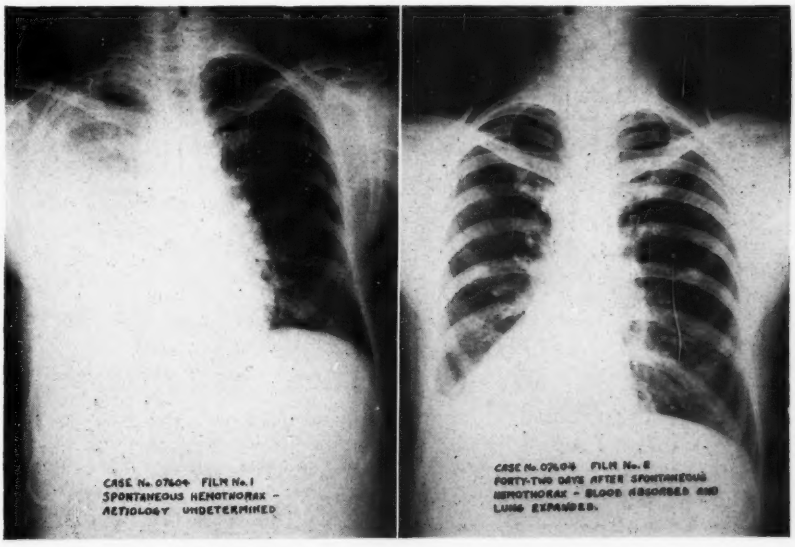
A. *B.*

Fig. 5.—*A*, Spontaneous pneumohemothorax. *B*, Radiograph shows re-expansion of lung in tuberculous pneumohemothorax after two and one-half years of pneumothorax.

Almeyda[81] cited a case of spontaneous hemothorax, and stated that the blood which settles on the diaphragm lacks fibrin. Birch[82] (1936) and Beatty[83] (1939) reported cases of hemothorax due to rupture of an alveolar cyst arising from scarred tuberculous foci. Davidson, Oxfd, and Simpson[84] reported a case of apical bullae and the tearing of adhesion which caused a hemorrhage. Perry and Camb[85] (1938) reviewed nineteen cases in the literature and reported two cases of their own. No evidence of tuberculosis was found in eight of the patients who died. In four autopsies of their cases a ruptured vascular adhes-

ion was found, and in the other no satisfactory explanation was found. Two cases of hemothorax are reported herein. Case 1 (Fig. 5, *A*) was tuberculous. Fig. 5, *B* shows a radiograph of the same patient after two and one-half years of pneumothorax treatment. Case 2 (Fig. 6, *A*) had a spontaneous hemothorax of nonspecific origin. Fig. 6, *B* is a film of the same patient forty-six days after blood had been absorbed and lung re-expanded.

CASE 1.—The patient, aged 24 years, a bank clerk, experienced a sudden pain in the right chest. Three hours after this attack he fainted. This was followed by considerable pain for several hours. He continued to work the next day and visited a physician, who taped the chest for pleurisy. Two days later x-ray view showed the right side of the chest two-thirds filled with fluid and one-third air. The patient was admitted to the hospital. A thoracentesis revealed the fluid to be blood. Guinea pig inoculation was positive for tubercle bacilli. The blood was gradually removed and replaced with air. The blood was totally removed in one month's time. Pneumothorax was continued for two years and the lung permitted to re-expand. Culture and gastric analyses were negative for tubercle bacilli.


CASE No. 07604 FILM No. 1
SPONTANEOUS HEMOTHORAX -
AETIOLOGY UNDETERMINED
CASE No. 07604 FILM No. 6
FORTY-TWO DAYS AFTER SPONTANEOUS
HEMOTHORAX - BLOOD ABSORBED AND
LUNG EXPANDED.

A. *B.*

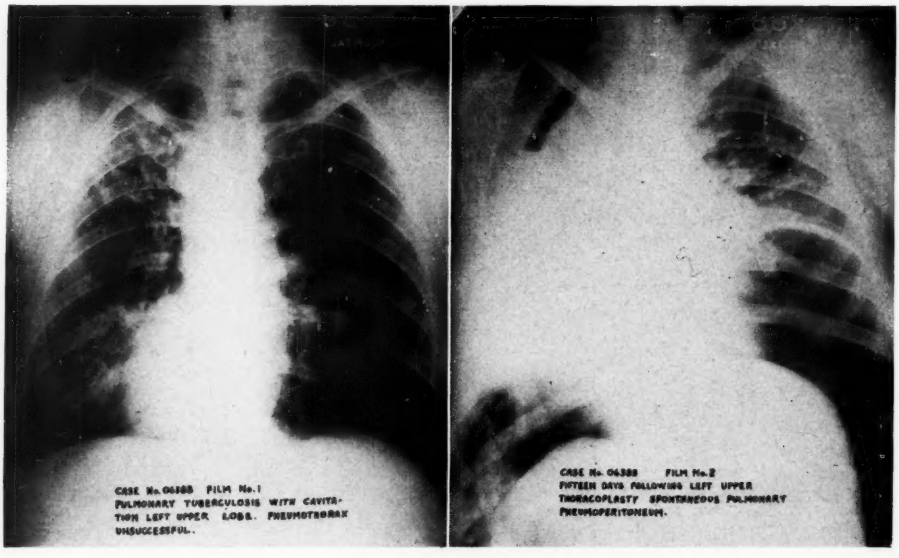
Fig. 6.—*A*, Spontaneous hemothorax. *B*, Forty-two days after hemorrhage. Lung re-expanded.

CASE 2.—The patient, aged 48 years, a housewife, developed sudden pain in the abdomen, and cramps and nausea. Complaints were referred to the abdomen, which caused one to suspect some involvement in the left kidney and abdominal region. Forty-eight hours later the patient complained of shortness of breath, which led to an x-ray examination. This revealed increased density in the left pleural space, and a partially collapsed lung. Marked secondary anemia was present. Thoracentesis revealed pure blood. No bacteriologic studies were made. Several hundred cubic centimeters of blood were removed, which gave the patient immediate relief. The remainder of the blood was absorbed. Three transfusions restored the blood picture to normal and the patient made an uneventful recovery.

SPONTANEOUS PULMONARY PNEUMOPERITONEUM

Spontaneous pulmonary pneumoperitoneum is an extremely rare condition. Indirect pneumoperitoneum, due to the flow of air from the pleural space into the abdominal cavity, is not too frequent. Indirect pneumoperitoneum has occurred following pneumothorax and during anesthesia, where excessive pressure has been the etiologic factor. Elrick,[86] Fishberg,[87] Stephani, Stephani, and Desboeuf,[88] and Banyai[89] have reported cases following pneumothorax. Eisen[90] in 1938 reported a case of spontaneous pneumothorax, mediastinal emphysema, and pneumoperitoneum which occurred during a closed ether anesthesia.

In 1944 Sidel and Wolbarsht[91] reported a case of indirect pneumoperitoneum, probably caused by a suspected temporary fistula which extended through the diaphragm into the abdominal cavity. This patient had pneumonia in the lower lobe of the left lung. The roentgenogram showed increased density in this area and a layer of air below the diaphragm. The patient recovered, and the air disappeared within two months.


CASE No. 06388 FILM No. 1
PULMONARY TUBERCULOSIS WITH CAVITATION LEFT UPPER LOBE. PNEUMOTHORAX UNSUCCESSFUL.
CASE No. 06388 FILM No. 2
FIFTEEN DAYS FOLLOWING LEFT UPPER THORACOPLASTY SPONTANEOUS PULMONARY PNEUMOPERITONEUM.

A. *B.*

Fig. 7.—*A*, Pulmonary tuberculosis with cavitation of left upper lobe. *B*, Spontaneous pulmonary pneumoperitoneum. Pocket of air at left apex is between scapula and chest wall.

The case herein reported (Fig. 7, *A*) was a patient with pulmonary tuberculosis, who had a left thoracoplasty (one stage, three rib section.) He recovered from the operation, but had discomfort from abdominal distention. On the fifteenth day the abdomen became markedly distended. An x-ray picture (Fig. 7, *B*) revealed pneumoperitoneum. This air was removed and patient was relieved. One week later he died of coronary occlusion. Autopsy showed

that a fistula probably existed from the lung through the diaphragm. The fistula was not demonstrated definitely. Adhesions above the diaphragm were densely adherent, and the omentum was adherent opposite the pleural adhesions on inferior surface of the diaphragm. No retroperitoneal or viscus break was observed.

The pneumoperitoneum in this case and the one of Sidel and Wolbarsht[91] was evidently caused by a direct connection from the lung through the diaphragm into the abdominal cavity. These cases demonstrate that a spontaneous pulmonary pneumoperitoneum does occur when a pathologic process creates an opening through the diaphragm. In the case reported here the fistula was valvelike, if the early abdominal distention was due to extraneous air and not flatus. This postoperative pneumoperitoneum (Fig. 7, *B*) occurred because of a pathologic process, even though the operation may have hastened the opening of a potential fistula.

CASE 1.—The patient, aged 61 years, a banker, white man, had been ill for one year with pulmonary tuberculosis. X-ray showed infiltration in the left lung with cavitation in the apex and a moderate amount of infiltration and fibrosis in the right upper lobe. An unsuccessful pneumothorax on the left side had been discontinued. He was referred for thoracoplasty. On Feb. 5, 1945, a posterior left thoracoplasty was performed. Recovery was uneventful, except for some distention of the abdomen, until the fifteenth postoperative day. Temperature elevated to 104° F., pulse rate 120, respiration 24. A diagnosis of pneumoperitoneum was made, and the patient was given relief by aspiration of air. He died of coronary occlusion one week later.

Autopsy Report.—The aorta was enlarged and atheromatous. The right ventricle was moderately dilated, and the wall contained two areas of healed infarction. The anterior coronary was calcified throughout, and part of the circumflex coronary was occluded. Considerable fibrosis and numerous healed tubercles were present in both lobes of the left lung. The cavity was collapsed. The lower lobe was adherent to the diaphragm, except in the anterior third. A few ounces of thick exudate were found in the costophrenic space. The omentum was plastered to the inferior surface opposite the dense adhesions on the pleural surface. An attempt was made to locate the fistula, but no definite opening could be demonstrated. The right lung was fibrotic and contained a few healed tubercles. The pleurae were adherent, except where a small quantity of fluid existed in the left costophrenic angle. There was no break in the peritoneum at the hiatus for the esophagus and aorta. The viscera were intact. The large bowel was distended markedly. The middle third of the transverse colon was adherent to the cecum.

SPONTANEOUS ATELECTATIC COLLAPSE

Spontaneous lung collapse caused by the presence of extraneous air is to be differentitated from the airless lung present in acquired atelectatic collapse. Pinner[92] believed that atelectasis cannot be produced by external pressure upon the lung. He concluded that if a lung under collapse therapy or after spontaneous pneumothorax is atelectatic, there is definite indication that some such complication as endobronchial occlusion or kinking has occurred. Atelectatic collapse may be acute or chronic, involving one or more lobes or segments of different lobes of the lung. As a rule, acute segmental and chronic lobular atelectasis occurs as a complication in pulmonary disease, rather than as a definite clinical entity, like massive collapse. Acute massive collapse occurs gen-

erally after abdominal operation within forty-eight hours, less frequently within a week.

According to Lee, Ravdin, Tucker, and Pendergrass[93] Mendelsohn produced massive collapse experimentally in 1841. These investigators, Coryllos and Birnbaum,[94] and Jackson and Lee[95] demonstrated bronchial blockade by viscid secretion to be the cause of massive collapse. Graham, Singer, and Ballon[96] reviewed all previous theories advanced up to the time Lee and associates[93] proved that simple obstruction was the primary etiology. Although Pasteur's[97] theory that diaphragmatic paralysis is the cause of massive collapse has been disproved, he described the original clinical condition in 1890.

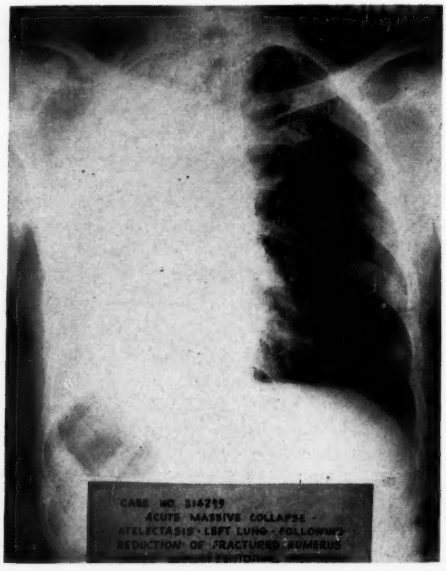


Fig. 8.—Acute, massive (atelectatic) collapse. Patient recovered after four bronchoscopic aspirations. (Courtesy of Blankenhorn and Felson, Cincinnati General Hospital.)

In massive collapse the early symptom is acute pain on the affected side, as in acute pleurisy. The pain is due to tension in the potential pleural space, and shifting of the mediastinum toward the involved lung. Besides pain, other symptoms are respiratory embarrassment, fever, and expectoration of blood-streaked sputum. Cyanosis and shock may be present. Physical signs consist of diminished breath sounds over the dull area, usually without râles. The heart apez beat is displaced toward the affected lung. The opposite lung is subjected to compensatory distention, which should be called physiologic emphysema. The x-ray view verifies the diagnosis with trachea and heart displaced toward the involved lung and the diaphragm elevated. Bronchoscopy is indicated. Carbon dioxide inhalation has been used both preventively and curatively. Moorman[98] in 1930, Schindler and Gnagi[99] in 1938, and Jacobson

and co-workers[100] in 1939 treated cases of massive collapse with pneumothorax. One case (Fig. 8, *A*) which was treated by bronchoscopy is reported.

CASE 1.—A white man, aged 45 years, a window cleaner, had a fracture of the right humerus, radius, and olecranon. Under Pentothal Sodium anesthesia a pin was inserted through the head of the humerus. On the second day he developed postoperative fever with sign of atelectasis (massive collapse.) This condition was treated with bronchoscopy on the third, sixth, eighth, and thirteenth day. A preoperative film was negative, except for moderate thickening of the left lower bronchi. Serial x-ray views showed gradual clearing of the entire left lung with exception of a little residual in the lower lobe.

SUMMARY

1. The continuity of interstitial, mediastinal, and subcutaneous emphysema is analyzed. Two cases are added to the few already recorded in the literature.

2. The etiology, diagnosis, and treatment of spontaneous collapse of the lung with air or blood is reviewed. Of the twenty cases of idiopathic spontaneous pneumothorax reported in this series, only three required treatment. In the three cases an irritant oil was used to initiate obliterative pleuritis. Twelve cases of spontaneous pathologic pneumothorax and two cases of nontraumatic hemothorax are reported.

3. Spontaneous pulmonary pneumoperitoneum with report of case is discussed.

4. Spontaneous lung collapse, caused by the presence of extraneous air, is differentiated from the airless lung present in acquired atelectatic (massive) collapse.

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A METHOD OF BRONCHIAL OCCLUSION FOR THE PREVENTION OF TRANSBRONCHIAL SPREAD DURING LOBECTOMY AND PNEUMONECTOMY: CLINICAL APPLICATION

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DURHAM, N. C.

IMPROVEMENTS in the methods for control of bronchial secretions during resection for suppurative pulmonary diseases have not kept abreast of the rapid progress in the development of surgical technique. In an attempt to answer such a need, an endobronchial balloon has been devised to occlude the bronchus to an affected lobe and thereby prevent the spread of purulent secretions. A résumé of the experimental approach to the problem has been published;[1] since August, 1946, this instrument has been applied clinically in 23 cases.

At least four different methods have been used in the past and are still being used in an attempt to solve this problem. These consist of tracheobronchial aspiration by catheter, mechanical blocking or isolation of one or more lobes, various positions for the patients with dependence on gravity control of the secretions, and different types of chemotherapy. The first method, that of tracheobronchial aspiration by catheter, undoubtedly has been employed much longer than the others and, in general, is the one method on which the greatest dependence has been placed. There is no doubt but that this method has its shortcomings. Bradshaw and O'Neill[2] in 1943 reported 76 cases of pulmonary resection for bronchiectasis with the use of this technique—there were 6 deaths directly due to contralateral spread. In a recent review of 125 cases of pulmonary resection performed at this hospital during the past ten years, it has been observed that ipsilateral and contralateral spreads occurred in 27, or 21.6 per cent. Of this group there were 5 deaths, all directly attributable to the spread. As in the cases of Bradshaw and O'Neill, tracheobronchial aspiration was the method routinely used.

There have been numerous attempts to solve this problem of contralateral spread by the use of mechanical devices. In addition to the instruments of Gale and Waters,[3] Archibald,[4] Magill,[5] Nosworthy,[6] and Crafoord,[7] there has come to our attention another device originated by Halton[8] of Liverpool, England. This consists essentially of a Magill endotracheal tube through which is inserted a fine hollow steel tube on the end of which is mounted an inflatable rubber sac. The balloon tube is clamped at predetermined levels at the proximal end of the Magill tube. According to Halton, the instrument could be inserted blindly with dependence on predetermined measurements of the tracheobronchial tree, or visually alongside a bronchoscope. No cases were reported in this original paper, and no further communication concerning the instrument has been found. All of these instruments have a similar basic construction; the unit is either rigid or semirigid, and the mouth of the patient is used

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as a fixed point. As will be discussed later, it would seem wise to avoid this if possible.

Various positions have been described as beneficial for these patients. Beecher[9] has recommended the Trendelenburg position and tracheobronchial suction by catheter. Blades and Graham[10] on the other hand were using the upright position in 1942; they readily admitted the dangers of shock with such a technique. Recently Overholt and associates[11] published a report on the use of the prone position and the preservation of the cough reflex through the use of local anesthesia; despite a significant decrease, the incidence of contralateral spreads was still 10 per cent.

The use of various drugs in the preoperative management of these patients has, within the last few years, opened a new avenue for the reduction of postoperative complications; it is readily apparent that the problem in question may be furthered to a solution but can never be completely solved through the use of chemotherapy alone. The recent reports of Findlay and Sweet[12] from Boston, and of Menefee and Fogel[13] from this clinic, plus many others, indicate that with the use of penicillin, particularly in aerosol form, the early mild cases of bronchiectasis can be carried through operation quite safely without fear of postoperative spreads. Although penicillin and streptomycin, in parenteral and inhalation forms, may reduce the amount of sputum in the cases of long-standing saccular bronchiectasis, chronic lung abscesses, secondary suppuration in bronchogenic carcinoma, and pulmonary tuberculosis, it has not seemed to be reduced to the level at which one could say that the danger of postoperative spreads is eliminated.

It is apparent from this discussion and from the numerous reports appearing in the literature, that there is still a need for some technique or instrument which will prevent the flooding of unaffected lobes with pus during operation.

Of the four methods of attack, it seemed apparent that improvements could best be made in the method utilizing instrumental bronchial occlusion. Such an instrument should meet certain qualifications. It must have no harmful effects; completely occlude the bronchus; remain securely in place during operative manipulations; have a wide margin of safety in its construction and operation, allowing complete control at all stages; and interfere as little as possible with anesthesia. In addition of these five points, it is essential that the instrument be inserted without trauma to the patient and that the operator know exactly where the instrument is placed. Furthermore, one should have no difficulty in removing the instrument when the bronchus is ready to be clamped.

The chief disadvantage of most of the instruments described has been their tendency to slip ont of place at inopportune times, plus difficulty in their insertion. Many of the others have lacked the essential point of being constructed so as to permit insertion and placement under direct vision. These factors could be answered by an instrument which utilizes the bronchus as a fixed point; in other words, the instrument would be flexible and thereby capable of being inserted through a bronchoscope.

These factors have been embodied in an instrument which has been developed at Duke Hospital, and which it is believed fulfills the necessary qualifications. The expected complications which have arisen in the transfer from the experimental animal to the patient have gradually been eliminated. In the first 3 cases in which we attempted to use the instrument, there were such gross errors in technique that the placement of the balloon was unsuccessful and since occlusion of the bronchus was not obtained because of these mistakes, which mostly concerned the instrument itself, these cases are not included in the present series reported.

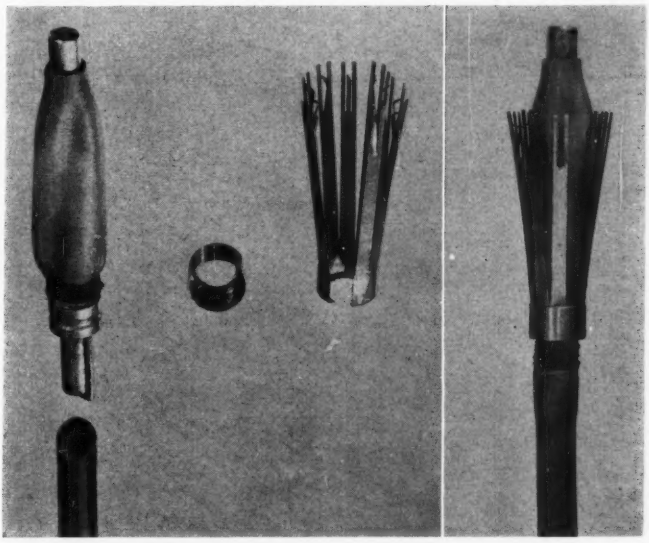

Fig. 1.—Endobronchial balloon, showing the essential parts and the assembled instrument.

DESCRIPTION OF INSTRUMENT

The instrument consists essentially of a small rubber balloon mounted on the end of a 3 foot length of No. 12 French double-lumen tubing. One lumen of this tubing passes to the balloon while the other continues through the balloon and ends in a small metal suction tip. Water under mercury pressure inflates the balloon. Dislodgment from the bronchus is prevented by a metal apparatus which encases the balloon and consists of several narrow staves which have recessed 1 mm. points (Fig. 1). The latter are embedded in the bronchial wall by inflation of the balloon, preventing its slipping back into the trachea. This holding apparatus is made in three lengths, corresponding to the diameters of the various bronchi where the balloon is placed The proximal end of the double-lumen catheter is connected to a small sleeve valve which maintains the balloon pressure while the bronchoscope is being removed. The remaining parts of the unit consist of a carrier tube for placement of the balloon, plus a mercury manometer and suction outlet. The latter are mounted on the side of a small table which serves also as a bronchoscopic table (Fig. 2). In order to maintain an airtight connection at the mouth of the patient, we use a face mask which has a small hole in one side through which is threaded the balloon catheter. When used for other purposes, the hole in the mask is closed with a plastic plug.

The functions of this instrument are twofold. It is designed to obstruct completely a bronchus to an affected lobe (or lobes), and thereby prevent the transbronchial spread of purulent material. By means of suction applied through and distal to the balloon, it also serves to produce atelectasis of the portion of lung to be resected (Fig. 3).

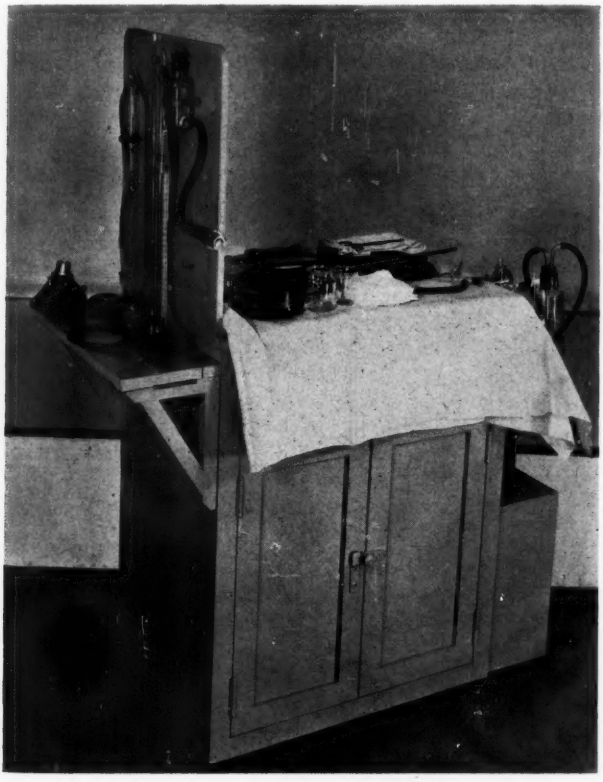

Fig. 2.—Movable table utilized as a bronchoscopy table, and as a mounting for the instrument panel.

The procedure followed in the present series of cases consists essentially of the following: Bronchoscopy is carried out on the patient following topical pontocaine anesthesia, which is applied to the trachea and bronchi as well as to the larynx and pharynx. The tracheobronchial tree is then thoroughly aspirated. Since the balloon projects a known distance of 1 cm. from the tip of the bronchoscope, the site of placement is easily localized. After this is done, pontocaine is applied directly to the site. The carrier tube containing the balloon is then inserted and the balloon released. Following inflation of the balloon to the desired pressure, the sleeve valve is turned off, disengaged, and the bronchoscope and carrier tube are removed. An endotracheal tube is then threaded over the balloon catheter and inserted, and the patient carried over into general anesthesia. The sleeve valve is recoupled as the patient is being inducted and the pressure in the balloon checked on the manometer. It is important that suction be applied distal to the balloon as soon as satisfactory anesthesia is obtained in order to prevent plugging of the aspirating tip by retained secretions. During the subsequent operation the pressure in the balloon is indicated and controlled by the mercury manometer. Following isolation of the bronchus the balloon is deflated and with-

drawn and the bronchus quickly clamped. The proximal portion of the bronchus is aspirated for any secretions which may have spilled out as the balloon was deflated. In the cases of bilateral disease, there is ample room in the endotracheal tube for passage of a suction catheter. A length of plastic tubing has been utilized which has slightly more rigidity than the usual rubber catheter.

It should be understood that this instrument is not intended to be used for an upper lobe resection; an instrument has been designed for this purpose, but thus far there has been no opportunity to test it.

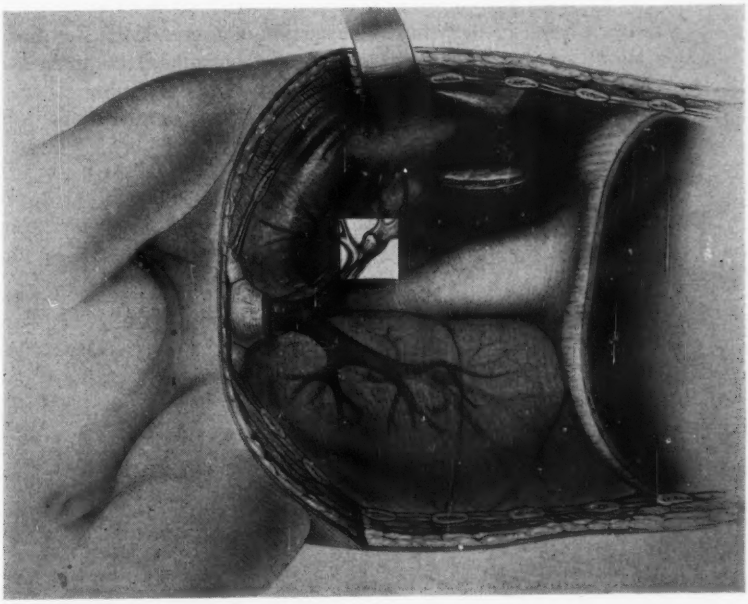

Fig. 3.—Drawing illustrating function of the instrument in obstructing the left lower lobe bronchus which communicates with a large abscess cavity.

REPORT OF CASES

During the past ten months this instrument has been used in a total of 23 cases. These consist of 10 lobectomies for bronchiectasis, 5 pneumonectomies for tuberculosis, 1 pneumonectomy and 3 thoracotomies for carcinoma, 2 pneumonectomies and 1 lobectomy for lung abscess. The remaining case was diagnosed lung abscess preoperatively, but was found to have a loculated empyema with a bronchopleural fistula.

Massive pulmonary suppuration was the primary indication for the balloon in 21 of the cases; this varied from 1 to 3 cups of sputum per day in nearly every patient. In 2 of the carinoma cases, the balloon was used solely for the production of atelectasis.

In all cases complete bronchial occlusion has occurred, evidenced either by atelectasis of the obstructed lobe, or lack of inflation under positive pressure anesthesia. There have been no obvious harmful effects in any of the patients. In the inoperable cases of carcinoma and in the patient with an em-

pyema, no postoperative complications have arisen due to the presence of the balloon in the main bronchus over a long period of time. In these cases, following removal of the balloon, the lung inflated very satisfactorily. There has been no interference with anesthesia at any time; on the contrary, it has been considerably aided by completely obviating tracheobronchial suction. The mechanics of operation and placement have proved quite simple. As a general rule, an average time of twenty minutes is all that is necessary from the beginning of the topical anesthesia to the completion of the placement of the balloon. Only one case has proved unsuccessful in insertion of the balloon; this was due to a combination of massive amounts of sputum and poor tracheobronchial anesthesia. The balloon has fulfilled its primary function of remaining securely in place in all except one case; in this instance there was extensive tuberculous infection of the bronchus at the site of placement of the balloon, and it was frankly suspected that the balloon would not remain in place, although it did so much longer than expected. There has been difficulty in removing the balloon in two cases at the time that the operator so desired. In one patient a sharply curved endotracheal tube prevented withdrawal of the instrument, and it had to be removed along with the balloon. In the second, the points of the holding apparatus were entirely too long and difficulty was experienced in releasing the balloon from the bronchus; this holding apparatus has since been discarded.

In this series of 23 cases, there has been one death on the operating table. This occurred in a young girl with massive unilateral tuberculosis and tuberculous bronchitis. Death occurred during hilar manipulation and shortly after removal of the balloon. There was no apparent reason for the death, and post-mortem studies could not be obtained.

In general, atelectasis has been produced satisfactorily and this has constituted a very definite advantage in allowing the surgeon considerably more operating space while maintaining expansion of the unobstructed lobe; however, in a few cases it has been only partial due to plugging of the aspirating tip by thick purulent secretions.

DISCUSSION

If the problem of preventing contralateral spread were merely one of obstructing a bronchus during the resection of an affected lobe, then this instrument should offer a satisfactory answer. As mentioned before, it fulfills all of the necessary qualifications: obstruction of the bronchus is complete; it remains safely in place despite operative manipulations; there is no interference with anesthesia; the mechanics of placement and operation are simple; and there are no harmful effects to the patient. However, through the continued use of this instrument we are becoming more cognizant of the fact that spreads may occur long before the operative procedure, may occur during preliminary bronchoscopy, and may even be confused with cases of pneumonia due directly to poor postoperative care. Consequently we are now forming a routine in operating on these patients in which the operation is postponed until late in the morning or the early part of the afternoon, postural drainage is enforced for

several hours preoperatively, adequate anesthesia is provided for bronchoscopy, and a rigid plan including forced coughing and endotracheal suction is followed in the postoperative period. We have found that it is particularly important that adequate anesthesia be provided for bronchoscopy. There is little reason why a patient should have paroxysms of coughing during bronchoscopy when the trachea and major bronchi can be anesthetized as easily as the larynx and pharynx. In the one patient mentioned previously, in whom the balloon could not be inserted because of massive amounts of secretions, the anesthesia was quite inadequate, coughing occurred repeatedly, and there was a definite although small, postoperative contralateral spread.

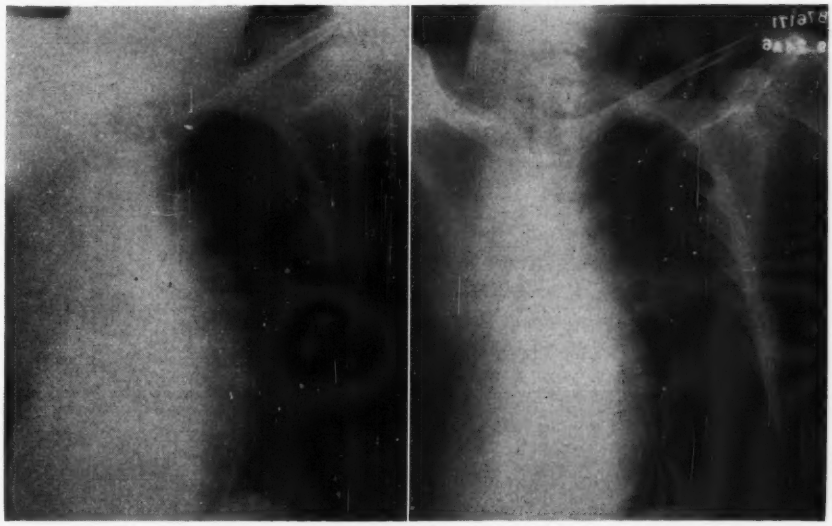

Fig. 4 (Case 1).—X-ray evidence of postoperative spread, which, twelve days later, showed beginning clearing. This spread may have occurred prior to insertion of the instrument.

The results in these cases are considered satisfactory, particularly when it is realized that this is a consecutive series of patients in which the balloon has been used.

Three examples of contralateral spread have occurred in the early cases of this series; fortunately they were all quite minimal and it was felt that they resulted from errors in technique which have since been corrected. One spread occurred in a pneumonectomy for tuberculosis with secondary bronchiectasis. The operation took place at 8:30 in the morning, the patient had not had adequate postural drainage, and bronchoscopy was difficult because of poor anesthesia. During operation bronchial occlusion was perfect, atelectasis was complete, and suction of the tracheobronchial tree was not necessary at any time. Despite this there was a definite small contralateral spread which slowly cleared on the patient's return to the sanatorium (Fig. 4). In the second case, a pneumonectomy for carcinoma, a sharply curved endotracheal tube prevented

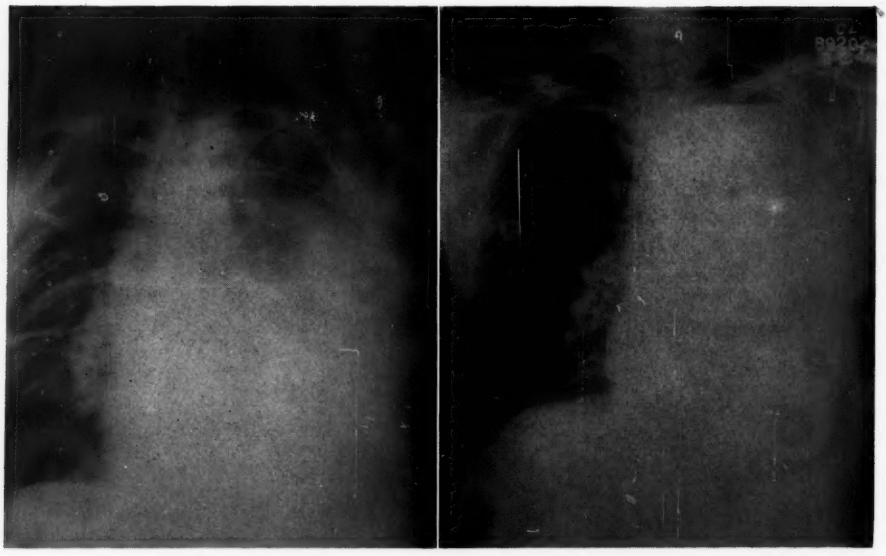

Fig. 5 (Case 2).—Very diffuse clouding of the right lung with clearing six days later. Bronchus was not clamped immediately on removing the balloon, and some spilling may have occurred.

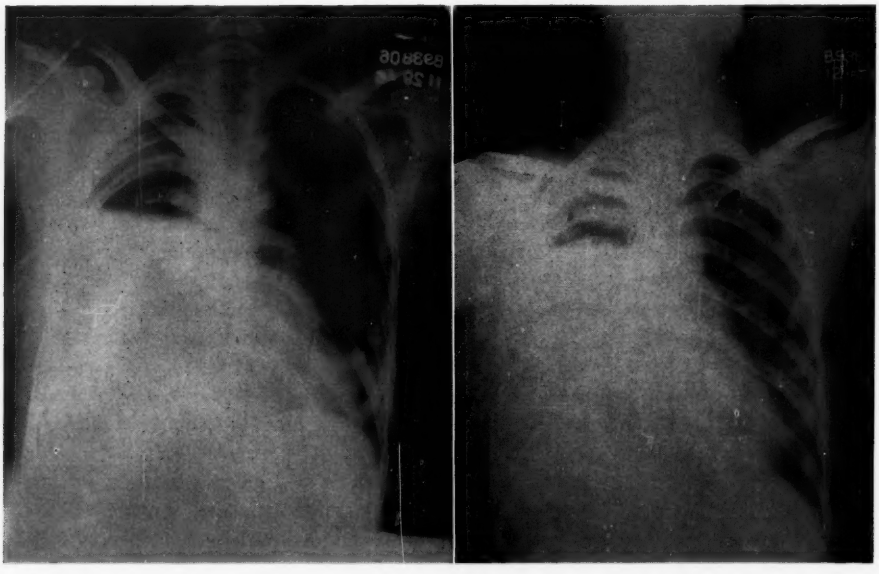

Fig. 6 (Case 3).—Very small area suggestive of spread in the left first interspace; this cleared considerably in one week's time. Bronchoscopy was difficult in this case.

the withdrawal of the balloon for several seconds, and finally had to be removed with the balloon. The bronchus was not clamped until the tube had been removed and consequently spillage of bronchial secretions may have occurred at this time (Fig. 5). The last example concerned a pneumonectomy for tuberculosis in which a small shadow appeared in the apex of the opposite lung on the third day, and showed definite clearing within one week's time (Fig. 6). During the operation the balloon functioned perfectly, and there was no break in technique to explain the spread.

This discussion is intended primarily as a report of the clinical application and the development of a technique for the use of an instrument which successfully occludes a bronchus during pulmonary resection. Any statistics on the reduced incident of spreads should be compiled from a large series of cases in which the use of this instrument is combined with the improved methods of pre- and postoperative care which have been discussed.

SUMMARY

Various methods for the prevention of transbronchial spreads during pulmonary resection for suppurative diseases have been used during the past few years. In an attempt to eliminate the faults apparent in these methods, an endobronchial balloon has been developed which has been successfully used in 23 cases. For the use of this instrument a technique has been developed which includes preoperative and postoperative care as well as the actual insertion and placement of the instrument.* It is hoped that the incidence of postoperative spreads may be greatly reduced by this method.

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*This instrument is to be available through the George P. Pilling & Son Co., Philadelphia, Pa.

PRIMARY RIB TUMORS

Survey of Literature and a Report of Seven Additional Cases

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Hedblom[1] in 1933 reported 291 cases of tumors of the bony chest wall, collected from the literature and from his own records. However, this number is not a true indication of the incidence of primary rib neoplasms since this group included inflammatory lesions, secondary growths, and neoplasia unclassified because of incomplete data.

In 1942 Sommer and Major[2] analyzed 81 proved cases of primary rib tumors reported subsequent to Hedblom's review, including 15 cases of their own. This number provides a more accurate indication of the incidence of these lesions.

The purpose of this paper is to list those cases described in the literature since the report of Sommer and Major, to review briefly clinical facts regarding rib tumors, and to report 7 cases of primary rib tumors treated at the State University of Iowa Hospitals. Table I shows these collected cases.

Primary tumors of the ribs present no clinical characteristics not found in similar tumors occurring elsewhere in the bony skeleton. Because of their location they do present certain diagnostic problems and surgical considerations which may well be reiterated.

The most important symptoms of rib neoplasms are pain and tumor. Pain persisting over a long period of time is probably a more reliable and constant finding than the presence of tumor since the growth may protrude little if any above the normal contour of the chest wall. Also, pain may develop before a mass is recognizable either clinically or roentgenographically. The pain is usually dull and boring in character and is not affected by respiration early in the progress of the disease, but later it may become pleuritic in character. Severe pain is most frequently associated with malignant growths. Fever, weight loss, dyspnea, palpitation, mild gastrointestinal distress, and pleurisy are other symptoms which occur rarely. In our series of 7 cases, pain was the initial symptom in 3 and was noted later in 2 additional cases; a tumor was the primary symptom in 3 and was noted later in two additional cases. One patient (Case 7) had pain but no tumefaction. Another, who was found to have a fibrous dysplasia of rib, had neither pain nor a mass.

The significance of trauma in the causation of neoplastic growths has been mentioned in practically every article concerning rib tumors.[1, 5-10] Just how

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important a role it plays is not yet clear. A history of local trauma was so frequently found in the reported cases that the traumatic factor may possibly be significant. Speed[20] stated that the majority of malignant tumors of the chest which appeared following trauma occurred in males and that the most common site was the anterolateral chest wall, the area most exposed to blows and falls. Hedblom found that 13 of 61 cases of sarcoma occurred in previously traumatized areas.[1] One case was reported wherein trauma occurred eighteen years prior to the appearance of a chondrosarcoma in rib.[6] The history of injury and subsequent distress during those many years was so clear cut that its contribution to the production of the tumor seemed difficult to deny. In a clinicopathologic conference at the Massachusetts General Hospital a case was presented in which an injury antedated a cavernous hemangioma of the rib by fifteen years. The history of a close relationship between the original trauma and the appearance of the tumor was such that coincidental occurrence seemed unlikely. One patient in our series (Case 2) developed a chest wall mass ten days after an injury to this area. The mass gradually enlarged and at operation six months later a fibrosarcoma was found.

Although the reports regarding trauma seem quite convincing, we are not ready to accept the theory of trauma as a causative factor in the development of bone tumors. More likely, a traumatic incident serves only to focus attention on a previously existing, asymptomatic pathologic lesion, whose presence was unknown before the injury occurred. We would therefore emphasize the fact that a previous injury does not permit a conclusion that persisting symptoms are resultant or insignificant. We feel that patients with pain and/or swelling which continues after trauma should be studied thoroughly for the presence of a tumor. Blake and Bradford have pointed out that too many cases are reported wherein roentgenograms have not been utilized, nor has there been an adequate physical examination in patients who complain of (a) persistent pain at the site of an injury over a period of time considerably longer than is required for healing of the injury to take place; (b) the appearance of a mass at the site of an old injury; (c) recurrence of pain at the site of an old injury after a lapse of months or years, when no immediate cause is apparent.[8] The fact that pathologic fractures may occur with minor trauma should be kept in mind. No complaints, such as those just mentioned, should be dismissed without complete x-ray studies even though the physical examination proved to be negative.

After one has made a diagnosis of disease in the thoracic cage, there are certain conditions which may be difficult for him to differentiate from rib tumors: These include tuberculous and pyogenic osteomyelitis of the spine or ribs. Syphilitic lesions must be considered as possibilities on occasion, although less frequently now than previously. Careful histories and routine blood and spinal fluid studies are important and will usually clarify the problem. Intrathoracic nerve tumors or tumors of the posterior mediastinum may occasionally be mistaken for bony chest wall lesions. Likewise, intercostal nerve tumors may simulate rib tumors clinically and roentgenographically. Case histories have

TABLE I

AUTHOR	CASE NO.	SEX	AGE	RIB SITE	INITIAL SYMPTOM	OTHER SYMPTOMS	DURATION	TREATMENT	RESULTS
					Chondrosarcoma				
Harper	1	F	60	7, 8 anteriorly	Pain	Dyspnea, tumor	10 yr.	Surgery	No recurrence at 9 mo.
Mayer	2	F	47	3 anteriorly	Tumor	Pain	3 yr.	Surgery	No recurrence at 5 yr. 3 mo.
Janes	3	M	60	9, 10, 11 anteriorly	Tumor	None	2 yr.	Surgery	No recurrence at 18 mo.
Janes	4	M	39	4 posteriorly	Pain & tumor	None	2 yr.	X-ray & surgery	No report
Janes	5	M	16	9 posteriorly	Dry cough	Pleural pain, shortness of breath	?	None	Died 1 mo. after onset
Janes	6	M	45	9, 10 anteriorly	Tumor	None	15 yr.	Surgery, local exc.	No recurrence at 2 mo.
Dorner & Marcy	7	F	51	11 laterally	Pain	Tumor	4 yr.	Surgery	Died 1 yr. later, metastasis?
Dorner & Marcy	8	M	56	6, 7 laterally	Tumor	Pain	3 yr.	Surgery, palliative	Died 3 mo. after op.
					Osteochondroma				
Janes	9	F	24	anteriorly	Tumor	None	8 yr.	Surgery	No recurrence at 12 yr.
Janes	10	M	51	1 anteriorly	Tumor	Pain	6 mo.	Surgery	Not stated
Bertola & Martinez	11	F	38	4 anteriorly	Tumor	None	1 yr.	Surgery	Not stated
Dorner & Marcy	12	F	33	2 anteriorly	Tumor	None	15 yr.	Surgery	No recurrence at 16 mo.
					Chondroma				
Janes	13	F	64	1, 2 anteriorly	Tumor	None	approx. 50 yr	Surgery	No recurrence
Janes	14	F	65	anteriorly	Tumor	None	30 yr.	None	Died of intercurrent infection

					Fibrosarcoma				
Blake & Bradford	15	M	20	6 anteriorly	Pain	Tumor; loss of weight; anorexia	2 yr.	None, inoperable	Died 21 mo. after onset
Dorner & Marcy	16	M	37	9, 10 anteriorly	Pain	Tumor	6 mo.	Surgery	Died 9 mo. after op.
					Hemangioma				
Mallory	17	M	34	6 posteriorly	Pain	Loss of sensation to heat & cold, left arm	15 yr.	Surgery	No report
Dorner & Marcy	18	F	22	9 posteriorly	Tumor	Pain	8 yr.	Surgery	No recurrence at 8 mo.
					Osteochondrosarcoma				
Ferrari & Lentino	19	M	—	9 laterally	Tumor	None	20 yr.	X-ray & surgery	No recurrence at 2 yr.
					Plasmocytoma				
Gonzales & Boggino	20	M	60	4, 5, 6, 7 laterally	Pain	Loss of weight headache, chills	3 mo.	X-ray palliative	Died 5 mo. after onset
					Osteosarcoma				
Luque	21	M	—	7 posteriorly	Tumor	None	2 yr.	Surgery	No recurrence at 2 mo.
					Lymphocytoma				
Estevez	22	F	27	9 anteriorly	Pain	Tumor, loss of weight	2 yr.	Surgery	No recurrence at 3 yr.
					Osteoid Osteoma				
Ponseti & Barta*	23	M	11	9 posteriorly	Pain	None	8 mo.	Surgery	No recurrence at 2½ yr.
					Fibrous Dysplasia of Rib†				
Lichtenstein & Jaffe	24	(No clinical data given)							
Schlumberger	Cases 25-54 (29 cases reported)								
Dorner & Marcy	55	F	21	6 posteriorly	None. Found on routine x-ray	None	Unknown	Surgery	No recurrence at 10 mo.

*Case 7 in our group, reported in J. Bone & Joint Surg. **29**: 767-780, 1947.

†These cases of fibrous dysplasia are listed with primary tumors of ribs chiefly because of their significance in differential diagnosis. In the 29 cases reported by Schlumberger, there was a wide range of diagnoses both by the roentgenologists and by the pathologists first studying the specimens. Almost all of these lesions were originally mistakenly diagnosed. Recognition of the true nature of these lesions is very important from a prognostic standpoint since the outlook is much better in these cases than in most bone tumors with which they are mistaken.

also been presented wherein rib tumors have been erroneously diagnosed and treated as simple contusions and fractures.[7, 9, 13] Lesions of the pleura, lung, and soft tissues of the chest wall (including aneurysm, cold abscess, encapsulated empyema, empyema necessitatis, and bronchiogenic carcinoma) have in some instances resembled bony tumors.

One of the most difficult problems, after demonstrating a bone neoplasm roentgenographically, may be the clinical differentiation between a primary and secondary growth. The ribs are involved by metastasis much more frequently than by primary growth. Ordinary methods of clinical examination may fail to disclose the source from which metastasis has occurred. Since secondary lesions in the ribs may become evident long before any symptoms are produced by the primary lesion, recognition of these facts is imperative.

A complete history and physical examination supplemented by the use of x-ray studies are the chief aids in diagnosing a rib tumor. In several of our cases, x-rays of most of the skeleton were taken to rule out polyostotic lesions. Likewise, special studies, particularly blood calcium, phosphorus, and phosphatase levels, were utilized to rule out diseases with secondary bone changes. Artificial pneumothorax and thoracoscopy after instilling air into the pleural cavity are diagnostic aids of probable importance in some instances. Some authors have suggested the use of aspiration biopsy, particularly to differentiate between primary and secondary tumors. This measure was not employed in our series. Exploratory biopsy remains a justified and essential procedure in those cases defying diagnosis by any other means and in which a positive diagnosis is necessary before management is instituted. Emphasis is made that this method is positively indicated in questionable cases rather than the frequently adopted plan of watchful waiting. It is apparent from the reports in the literature and from a review of our own cases that in too many instances there is a great delay between the onset of symptoms and the removal of the lesion. This is due in some cases to an unjustified wrong diagnosis and in others to a failure to identify lesions in their premalignant stage. For example, chondromas and osteochondromas are now recognized as being either potentially malignant from their onset or as having a tendency to become malignant later, and they demand adequate treatment at the time they are first noted.

The definitive treatment of primary rib tumors is no longer a controversial matter. There is little evidence that irradiation therapy is of real value. Except for two giant-cell tumors which responded favorably and a similar lesion apparently cured by radium therapy, there are no reports since 1933 of successful treatment by this means. Early and wide excision is the treatment indicated. By wide excision is meant removal of the rib including the periosteum and the underlying pleura well beyond the lesion along with the adjacent muscles (intercostals, chest wall, diaphragm, etc.). Not infrequently, reports state that the pleura has been stripped from a rib tumor rather than being widely excised. This was true in several of the earlier State University of Iowa Hospital cases, and in one instance (Case 1) we believe this fact was likely responsible for a recurrence. Entire removal, particularly of the first rib, for

a malignancy involving any portion of it, is advocated by Fort,[5] since partial resection in this extremely vascular area is frequently followed by early infiltration and rapid general metastasis. (In our Case 6 we removed not only all of the ninth rib and the underlying pleura and the eighth and ninth intercostal muscle bundles, but also a segment of the eighth rib in the region of the mass so as to facilitate wider excision. The microphotograph of this case [Fig. 6, *D*] shows the close relationship of the tumor capsule and the pleura. Obviously, if stripping of this tumor from the pleura had been attempted, complete removal would not have been accomplished).

In the following cases of primary rib tumors the patients have been operated upon at the State University of Iowa Hospital from 1934 to September, 1946.

CASE REPORTS

CASE 1.—F. T. G., a white woman, first entered the State University of Iowa Hospitals in 1934 at the age of 51 years, with a complaint of a mass on the lower left part of the chest of twenty years' duration. On physical examination there was a hard, slightly nodular, nontender mass measuring 15 by 10 by 10 cm. in the left posterior axillary line lying between the tenth and twelfth ribs. The mass was excised along with a fairly generous segment of the eleventh rib, from which it arose. The operator's note included the statements: "The mass peeled out quite readily, the only technical difficulty encountered being separating the thinned out pleura which had been displaced by the inward growth of the tumor."

Pathology.—Gross Description: The mass measured 15 by 11 by 10 cm. To it was attached a rib 3.5 cm. in length. The tumor appeared to rise from the end of the rib. The cut section revealed chiefly cartilage, but a few areas of bone were seen.

Microscopic Description (Fig. 1, *A*): Microscopic slides showed neoplastic cartilage, pleomorphic in appearance. In some areas the cartilage was lobulated and the cartilage matrix was dense. Here the cartilage cells tended to be large, ovoid, and irregularly distributed in the matrix. The matrix in some areas showed irregular focal calcification. In other areas the tumor was composed of much more embryonal cartilage; here the matrix was watery in appearance and devoid of calcification. Cartilaginous lacunar outlines were difficult to ascertain and the individual cartilage cells were more pleomorphic. Many of them were spindle-shaped and had long tapering cytoplasm. No mitotic figures were seen.

The diagnosis in 1934 was "chondroma of rib." However, because of the evidence of a mixture of mature and embryonal cartilage, we would now call this tumor a low-grade chondrosarcoma.

This tumor *did* recur. On the patient's second admission to the hospital in 1945, a recurrence which she had noted for the past two years was excised. This arose subjacent to the old operative scar.

Pathology.—Gross Description (Fig. 1, *C*): A 7 by 7 by 6 cm. well-circumscribed mass of cartilaginous tissue surrounded by a rather thick pseudocapsule was found. Upon sectioning, the tumor tissue bulged beyond its pseudocapsule.

Microscopic Description (Fig. 1, *D*): There were several sections of pleomorphic neoplastic cartilage similar in appearance to the original tumor. In these sections, however, a more immature type of cells and matrix predominated. Invasion of the pseudocapsule was seen in one area. There was minimal matricial calcification. No endochondral ossification was seen.

The patient's third admission was in September, 1946, in the department of otolaryngology, because of a mass in the left side of the mandible which had been noted two months previously. A diagnosis of chondrosarcoma was made and, following biopsy, the left external carotid artery was ligated and the mandible was excised. The patient suffered secondary shock and died about eight hours postoperatively. No autopsy was performed. The pathologic diagnosis was chondrosarcoma of the mandible.

Pathology.—Microscopic Description (Fig. 1, *B*): This tumor was similar in appearance to the previous specimens. As in the recurrent mass removed from the rib, this tumor was composed predominantly of fairly immature hyaline cartilage. However, there was fairly conspicuous calcification in some portions of the matrix. In sections of the same tumor removed at preliminary biopsy, one could see invasion into the buccal submucosa. The invading portion of the tumor was distinctly primitive in appearance.

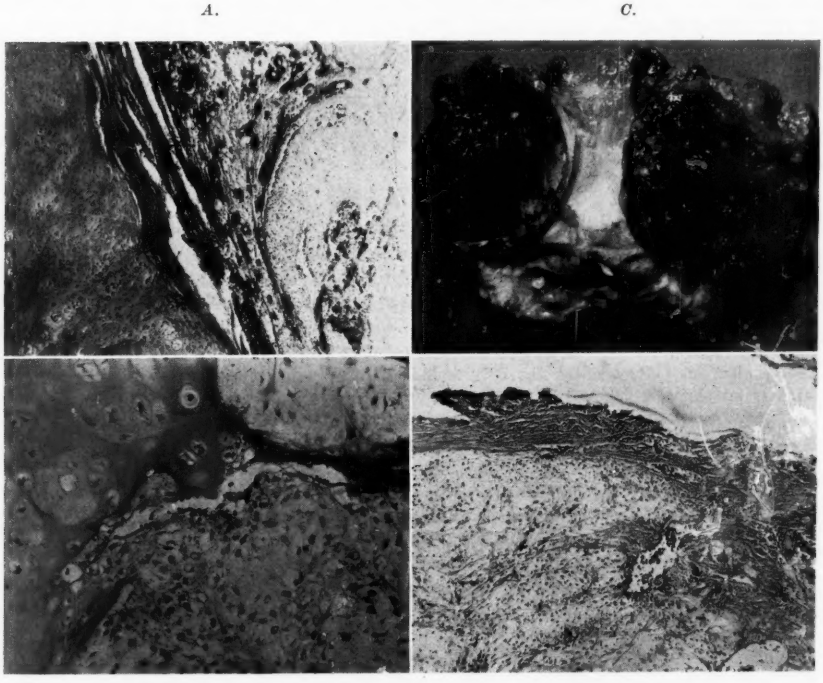


Fig. 1 (Case 1).—*A*, Photomicrograph (low power) of chondrosarcoma of rib showing pleomorphic character with watery cartilage matrix to the right of the connective tissue septum and fairly dense matrix with more mature cartilage cells to the left of the septum.

B, Photomicrograph (high power) of chondrosarcoma of mandible showing two lobules of primitive cartilage adjacent to more mature partially calcified hyaline cartilage. Note resemblance to tumor of rib above.

C, Photograph of recurrent chondrosarcoma of rib. The dense white band in the center is the pseudocapsule which has been laid open revealing the everted friable hyaline cartilage.

D, Photomicrograph of recurrent chondrosarcoma of rib shown above, showing immature neoplastic cartilage invading the tumor's pseudocapsule.

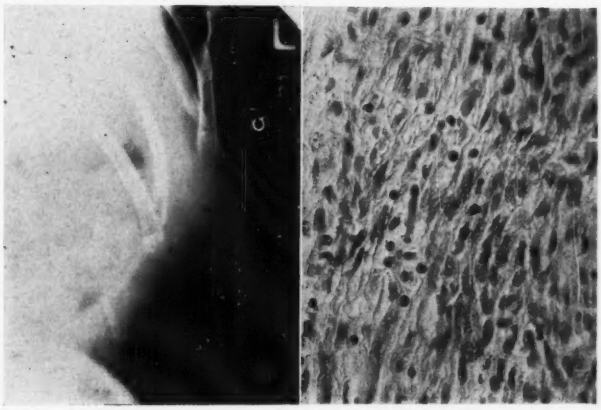
Discussion.—We feel that the first operation was inadequate. The overlying muscles, pleura, and intercostal muscles should have been widely excised. In resecting any tumor of rib it should never be stripped from its underlying pleura. The recurrence in this case may well have been due to this mistake in technique.

Regarding the lesion of the mandible—one may speculate as to the relationship of this tumor and those of the rib. We feel that this probably was not a metastasis but rather a new tumor. The patient may have had enchondral rests and for some reason these developed as chondrosarcomas. On the other

hand, the similarity of the microscopic pictures might lead one to believe that the lesion in the mandible was a metastasis which might have been avoided had an early wide excision of the primary tumor of rib been carried out.

CASE 2.—J. H. a 37-year-old white man, was admitted to the State University of Iowa Hospitals on May 12, 1938. He complained of a mass on the left side which had appeared ten days after a fall on the chest in November, 1937. Surgical removal had recently been attempted at another hospital but had failed. Examination revealed a mass, 12 by 8 by 6 cm. in size, which occupied the region of the costal margin on the left and was adherent to the ninth and tenth ribs (Fig. 2, *A*). A block resection of the mass was carried out, removing the eighth, ninth, tenth, and eleventh ribs together with their costal cartilages and the underlying endothoracic fascia and pleura.

Pathology.—Gross Description: The mass measured 12 by 5 by 4 cm. One rib disappeared into a hard mass measuring 7 by 5 by 5 cm. This mass was quite well encapsulated and on cut surface was composed of translucent grayish tissue.



A. *B.*

Fig. 2 (Case 2).—*A*, X-ray view of fibrosarcoma of rib showing destruction of rib and the soft tissue mass. *B*, Photomicrograph of fibrosarcoma of rib showing spindle cells and numerous mitotic figures.

Microscopic Description (Fig. 2, *B*): Sections showed interlacing bundles of neoplastic cells which were spindle-shaped and had elongated oval nuclei. These cells tended to run in parallel direction, forming loosely packed whorls and long sheets and interlacing bands. Variation in cell size and depth of staining was frequent. Mitotic figures were abundant. Invasion of bone and adjacent soft tissue was widespread. There was considerable periosteal elevation with formation of immature subperiosteal new bone. In areas scattered throughout the periphery of the tumor there was nonneoplastic new bone formation. Some of this was represented by immature osteoid tissue.

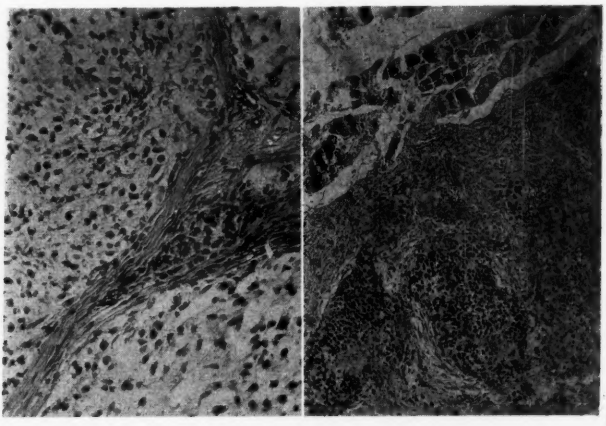
The diagnosis of fibrosarcoma of rib was made.

The patient resumed work as a sawmill engineer two and one-half months after operation and worked for a period of about five months. He died nine months after operation with widespread metastases as suggested by a letter received shortly after his death.

CASE 3.—C. W., a 56-year-old white man, was admitted to the State University of Iowa Hospitals on Jan. 21, 1938, complaining of a growth in the right chest wall for the past three years. The mass together with the sixth and seventh ribs had been resected two years previously at another clinic. The diagnosis had been "grade I fibromyxochondrosarcoma

of sixth and seventh ribs.'' Recurrence was noted six months later, however eighteen months elapsed before the patient was referred to the State University of Iowa Hospital. On Jan. 29, 1938, after preoperative pneumothorax, exploratory thoracotomy was performed. The lesion was found to be inoperable because of metastasis to the pleura and to mediastinal lymph nodes; therefore, a palliative excision was carried out. The pathologic diagnosis was osteochondrosarcoma, recurrent. He was discharged on the thirty-fourth postoperative day and died two months later.

Pathology.—Gross Description: The specimen consisted of an irregular, lobulated, nodular mass measuring 20 by 6 cm. This mass was soft in some areas and bony hard in others. Portions of rib could be seen here and there protruding from the mass. One area was cystic and contained yellowish, thick, viscid material. The mass was partially encapsulated. On cut section it was composed of irregular nodular masses of grayish-white tissue intermingling with fibrous tissue. There were areas of necrosis, hemorrhage, and calcification.



A. *B.*

Fig. 3 (Case 3).—*A*, Photomicrograph (high power) of recurrent chondrosarcoma of rib showing immature cartilage with connective tissue septa. *B*, Photomicrograph (low power) of recurrent fibrosarcoma of rib showing invasion of muscles of the chest wall.

Microscopic Description (Fig. 3, *A*): Sections showed lobulated masses of neoplastic hyaline cartilage tumor, surrounded by a dense fibrous tissue stroma separating this into lobules. These fibrous tissue septa were in many places dense, broad, and occasionally calcified. The cartilage neoplasm varied considerably in cellularity but predominantly was more cellular than normal hyaline cartilage. In the cellular areas the matrix was predominantly light-staining and devoid of calcification. These areas frequently appeared frankly myxomatous. In other areas the matrix was sparsely cellular and showed varying degrees of calcification. This calcification was the site of ossification in many areas. The ossification was predominantly by direct osteoblastic apposition. The osteoblasts and the bones thus formed did not appear neoplastic but were a reactive response to the calcified material. There were small areas where the neoplastic cartilage cells were distinctly pleomorphic and varied considerably in size, shape, and depth of staining. In one section lobules of neoplastic cartilage were seen pushing into skeletal muscle. A thin barrier of flattened connective tissue separated the two (Fig. 3, *B*).

Diagnosis was chondrosarcoma of rib.

CASE 4.—D. C., a 33-year-old white woman, entered the State University of Iowa Hospitals on Feb. 11, 1946, complaining of a painless mass in the region of the left second costochondral junction of fifteen years' duration. It had been increasing in size for some

time (Fig. 4, *A* and *C*). On Feb. 13, 1946, the mass was excised, including cartilage and rib, well away from the gross tumor. Included in the block resection were the adjacent intercostal muscles and underlying pleura. The defect of the chest wall was closed with the pectoralis minor muscle.

Pathology.—Gross Description (Fig. 4, *B*): The specimen consisted of a section of rib 7.5 by 4 by 4 cm., to which was attached intercostal muscles. The central portion was greatly widened and was composed of lobulated masses of cartilage. One surface was covered by shiny pleura. On section the lobulated masses were seen to be composed of dense cartilage. The overlying pleura was thin but intact. The attached tissues appeared to be displaced rather than invaded.

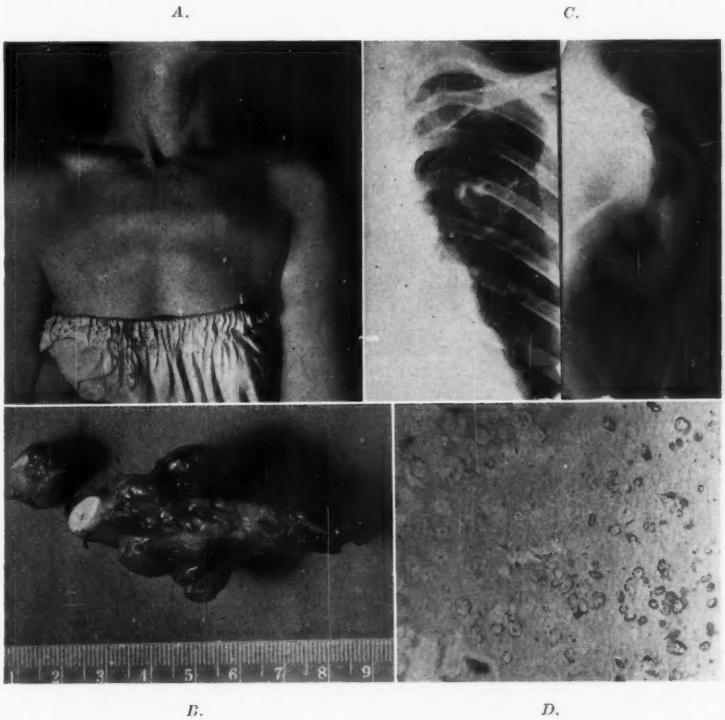


Fig. 4 (Case 4).—*A*, Photograph of patient showing tumor caused by osteochondroma arising at the left second costochondral junction. *B*, Osteochondroma of second rib with attached intercostal muscle and pleura. *C*, Posteroanterior and oblique x-ray views of osteochondroma of left second costochondral junction. *D*, Photomicrograph of osteochondroma of rib showing areas of calcification in relatively mature hyaline cartilage.

Microscopic Description (Fig. 4, *D*): Sections taken from various areas in the periphery revealed varying degrees of cartilage maturity. In some areas there was fairly mature hyaline cartilage with disorderly calcification and cartilage cell column arrangement. There were numerous scattered focal areas of senescence and calcification. Sluggish endochondral ossification was seen. A few areas in the perichondrium showed more immature cartilage with multifocal areas of proliferation. However, this did not appear to be invasive in nature and the appearance was consistent with that of a benign cartilaginous tumor.

Diagnosis was osteochondroma of the second rib on the left.

The patient's postoperative course was uneventful. A recent follow-up report from the physician for whom the patient works states that she is well at the present time.

Discussion.—Although this was diagnosed as a benign lesion, we feel that when osteochondromas of ribs arise they should be removed early by block resection rather than "watching them for any change" as had been done in this case for fifteen years.

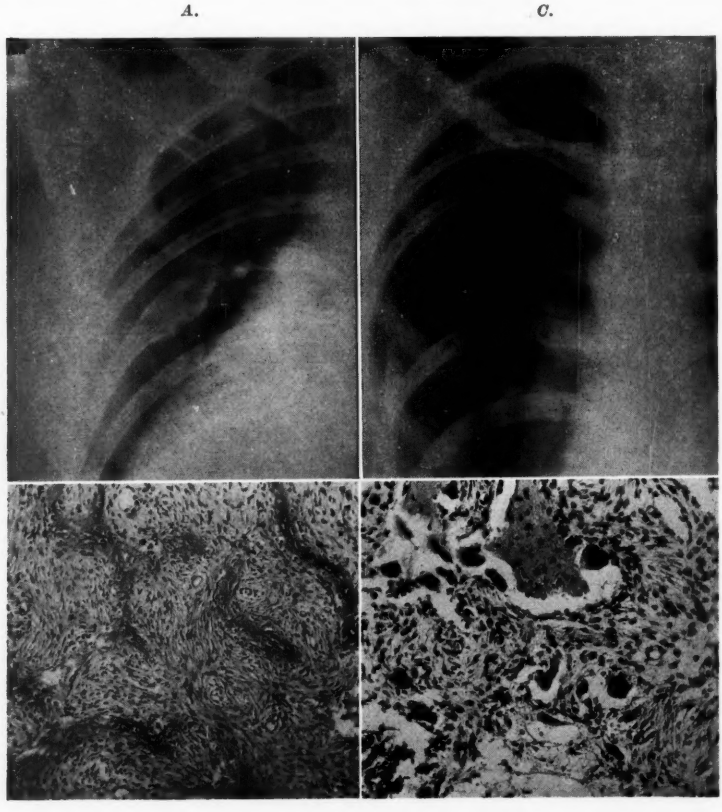


Fig. 5 (Case 5).—*A*, Preoperative x-ray of patient with fibrous dysplasia of left sixth rib. *B*, Photomicrograph (low power) of fibrous dysplasia of rib showing a fairly typical area of young fibrous tissue in marrow cavity containing delicate osteoid trabeculae. *C*, Postoperative x-ray. *D*, Photomicrograph (high power) of fibrous dysplasia showing multinucleated giant cells in cellular fibrous tissue and in Howship's lacunae.

CASE 5.—P. M. O., a 21-year-old white woman, a nurse, incident to a routine chest roentgenogram, was found to have a tumor of the left sixth rib (Fig. 5, *A*). She had no complaints. Laboratory studies revealed the blood and urine to be negative. The Wassermann was negative. Blood calcium, phosphorus, and phosphatase studies were within normal limits. X-ray films of the pelvis, both femurs and knees, both shoulders, mandible, and spine showed no evidence of bony disease. On July 7, 1946, a block resection of the involved rib was carried out, extending more than 2 cm. beyond the palpable tumor on each side and including the adjoining intercostal muscles and the underlying pleura. The patient's postoperative course was uneventful. Recent follow-up examination and x-ray films show no evidence of recurrence.

Pathology.—Gross Description: The specimen was a 7.5 cm. segment of rib. In the 4.5 cm. of one extremity the medullary canal was dilated from the normal width of 2 mm. to 6 mm. At one extremity of this widened portion was a softened, round, translucent area. The trabeculae of bone were condensed around this mass.

Microscopic Description (Fig. 5, *B* and *D*): The marrow space of the rib was replaced by masses of young fibrous tissue appearing in whorls and interlacing bands. In some areas there was direct metaplasia to bone in this cellular fibrous tissue. This bone was seen in the form of slender delicate irregular trabeculae. Many of these were represented by immature osteoid tissue which blended into the young fibrous tissue. The conspicuous osteoblastic appositional bone formation seen in osteoid osteoma was lacking. In some areas there were fairly numerous multinucleated giant cells lying in apposition to reactive new bone and in the cellular fibrous tissue. Some of these were lying in large Howship's lacunae. No blood pigment or lipoid-laden cells were seen.

Diagnosis was fibrous dysplasia of rib.

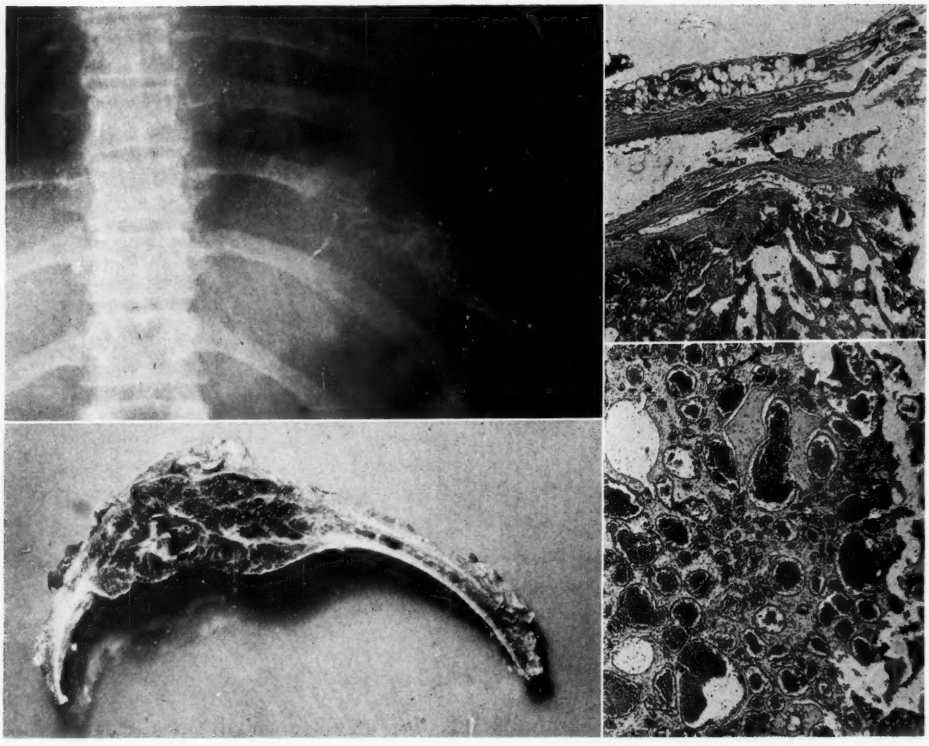
Discussion.—This case is quite typical of fibrous dysplasia of a single bone. As in 12 of 29 cases reported by Schlumberger, this case presented no complaints; it was found on routine x-ray of the chest. The histologic picture is similar to the changes in the "Albright syndrome." However, this patient presented no abnormal pigmentation of the skin nor any endocrine dysfunction. Lichtenstein and Jaffe, Schlumberger and others have described this lesion in considerable detail. The prognosis is good.

CASE 6.—C. S., a 22-year-old white woman, was admitted to the State University of Iowa Hospitals on Sept. 5, 1946. In 1938 she had noted a swelling which she described as being about "one-half the size of a hen's egg" in the region of the left ninth rib posteriorly. This was sore to pressure. There was never any local redness nor systemic reaction. The lump lasted about one week and then it and the associated symptoms disappeared. Approximately three weeks before admission to the State University of Iowa Hospital the patient again developed a sharp pain and soreness in this region. The local doctor took x-rays and noted a tumor of the left ninth rib (Fig. 6, *A*). He then referred her to the State University of Iowa Hospital.

Physical examination revealed a firm mass on the left ninth rib posteriorly, about 3 by 3 by 3 cm. in size, extending from the posterior axillary line posteriorly about 4 cm. It was not tender and there was no local erythema. Laboratory studies revealed the urine to be negative. Blood counts, and blood calcium, phosphorus, and phosphatase determinations were within normal limits.

On Sept. 6, 1946, a block resection of the entire left ninth rib including adjacent intercostal muscles and underlying pleura was carried out. A segment of the eighth rib was also removed to facilitate wider excision. The postoperative course was complicated by hemothorax which responded satisfactorily to thoracenteses. Recent follow-up reports from the patient's referring physician state that she is in perfect health. Roentgenograms show no evidence of recurrence.

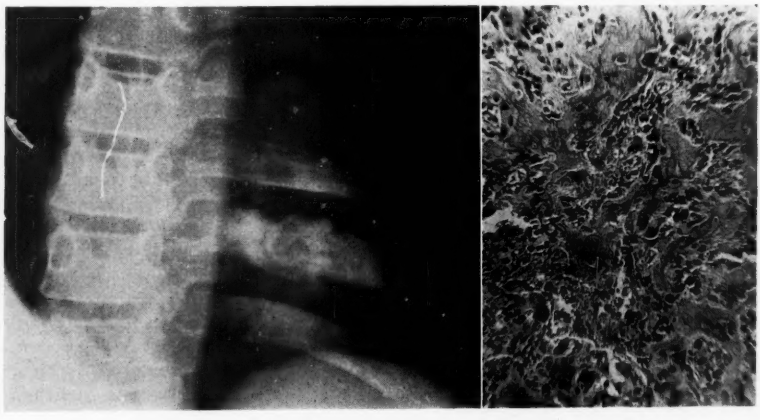
Pathology.—Gross Description (Fig. 6, *B*): The specimen consisted of a segment of rib measuring 16 cm. in length. Near the vertebral end of the rib the normal structure was replaced by an intrinsic tumor which measured 4.5 cm. in its greatest diameter. In the other diameter it was 4 cm. Its length along the course of the rib was 8 cm. The tumor was fusiform in shape. On external inspection there was no gross evidence of the tumor breaking through the parietal pleura or the intercostal fascia. However, in one spot there was a short tear in the parietal pleura which may have extended into the tumor. After sectioning the rib in longitudinal direction after partial fixation in formalin, it was seen that there was only a shadowy outline of the pre-existent osseous structure of the rib. This outline showed some expansion and irregular replacement with soft friable red tumor. Peripheral to the outline of the rib structure, there were radiating spicules of rather friable new bone



A. *C.*

B. *D.*

Fig. 6 (Case 6).—*A*, X-ray of hemangioma of left ninth rib. *B*, Photograph of rib involved by hemangioma after partial fixation in formalin and longitudinal sectioning. *C* and *D*, Composite photomicrograph of hemangioma of rib. Note the very thin capsule separating the tumor from the pleura.



A. *B.*

Fig. 7 (Case 7).—*A*, X-ray, view showing osteoid osteoma of left ninth rib. *B*, Photomicrograph of osteoid osteoma of rib.

in the so-called "sunray" appearance. In some areas there was more of a "honeycombed" appearance and the interstices were filled with this same rather mushy red tumor tissue which grossly resembled hemangioma. At the vertebral borders of the tumor extending in nodular manner under the parietal pleura the tumor appeared to be composed more largely of soft tissue with less reactive new bone. One margin of the tumor was composed of rather homogeneous firm yellow material. At this place it was tightly attached to overlying intercostal fascia.

Microscopic Description (Fig. 6, *D*): In all the sections decalcified there was evidence of hemangiomatous tissue which was invading and replacing the normal bone and marrow. Large blood-filled venous cavernae lined by venous endothelium varied greatly in size. These were present between active and sclerosing trabeculae of bone. The supporting structure of the vascular elements was composed of cellular connective tissue and a reticulum containing some interstitial fluid and hyalinized tissue. These supporting cells contained no mitoses. The supporting tissue contained some lymphocytes and some extravasated red blood cells. Bony trabeculae showed some destruction and irregular formation with irregular lamellae and cellular arrangement. The periosteum was for the most part intact about the cortical limits, but one area suggested outgrowth through periosteum. No extensive signs of invasion or malignant tendencies were present in any sections. In no place did the tumor show invasion of pleura, but in some areas it was separated from the pleura by only a very thin capsule (Fig. 6, *C*).

Diagnosis was hemangioma of the rib.

CASE 7.*—W. W., Jr., a 11-year-old white boy, was admitted to the State University of Iowa Orthopedic Clinic on Sept. 7, 1944, with a complaint of pain in the region of the left ninth rib for the past seven to nine months (Fig. 7, *A*). On Nov. 21, 1944, a subperiosteal resection of the proximal 2½ inches of the left ninth rib was carried out. The clinical diagnosis was enchondroma or osteoid osteoma. The last follow-up examination on April 11, 1945, showed no recurrence.

Pathology.—Gross Description: There were three pieces of bone representing the proximal portion of the ninth rib. There was a tumor 1 inch long and ⅝ inch thick, covered by apparently normal cortex. Inside there was a mass of sclerotic, whitish irregular bone. No cartilage could be seen macroscopically.

Microscopic Description (Fig. 7, *B*): Two slides were studied. The tumor mass was made of osteoblastic tissue with abundant bone formation. In the center of the mass this bone was quite dense but at the periphery the osteoid was very immature, irregular, and the osteoblasts were quite anaplastic in appearance. In the cellular peripheral areas many of the osteoblasts had abnormally large and hyperchromic nuclei. Scattered mitoses were present. This area resembled osteogenic sarcoma and in one area the cortex of the rib was being invaded.

Diagnosis was osteoid osteoma, unusually cellular—anaplastic at margins.

Discussion.—The clinical course together with central sclerosis gives the typical picture of an osteoid osteoma and it is felt that the lesion is probably classified in this group. Because of the extreme anaplasia and cellularity peripherally it is felt that the prognosis should be somewhat guarded.

Three patients (Cases 4, 5 and 6) were operated upon by one of the authors (R.A.D.) with the policy of wide excision being carried out. These three, as well as one other (Case 7), are still alive and well; the others have died. We would not imply that the wide excision in Cases 4, 5, and 6 is responsible for the patients' survivals but wish only to emphasize our present policy of this type of therapy for all primary tumors of rib.

*This case was reported with a series of osteoid osteomas in J. Bone & Joint Surg. 29: 767-780, 1947.

SUMMARY

1. The literature regarding primary rib tumors has been briefly reviewed. Fifteen cases since a comprehensive review by Sommer and Major in 1942 are listed. Seven additional patients operated on at the State University of Iowa Hospital are reported upon.

2. Pain and tumor are the most frequent complaints.

3. Trauma as a possible etiologic factor is discussed. We believe that pain and/or a tumor persisting after trauma should not be considered lightly but should be investigated thoroughly as possibly being due to a rib tumor.

4. The diagnosis of primary rib tumor is discussed in some detail.

5. We believe that early wide excision including resection of periosteum, underlying pleura, and adjacent muscles is the treatment of choice for all primary rib tumors.

Addendum.—Since this paper was submitted for publication another patient with a primary rib tumor has been treated. The patient, a 33-year-old white man, had had a painful tumor of the left tenth rib for at least one year. The mass had been biopsied in December, 1947, and was found to be an osteochondromyxosarcoma. X-ray treatment was given. On March 29, 1948, at the Iowa Methodist Hospital, a radical excision, including a portion of the diaphragm which was invaded by the tumor, was carried out. When last checked on July 30, 1948, the patient appeared well.

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CAVERNOUS ANGIOMA OF THE RIGHT LUNG

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THE number of cases of cavernous angioma (fistula arteriovenosum, aneurysma arteriovenosum) of the lung which have been recorded is still so small that the history, diagnosis, and treatment of such a case are worthy of presentation.

CASE REPORT

H. M. S., a boy, was born Feb. 4, 1937, one month prematurely. He was cyanotic at birth, apparently by strangulation. The cyanosis soon diminished, although the hands remained somewhat blue in color.

At the age of 10 months, the patient developed whooping cough and pneumonia, and in 1939 he had scarlet fever. In July, 1939, on x-ray film, a round, speckled shadow was found in the lower part of the upper lobe of the right lung.

Since the Mantoux reaction was positive, the boy was considered to have tuberculosis and was sent to a sanatorium for one year. Cyanosis at that time was very slight, but there was marked clubbing of the fingers and toes.

During October, 1944, the patient had several hemoptyses, sometimes more than 100 c.c. being expelled. Furthermore, he developed convulsive movements in the left side of the body. At this time he was distinctly cyanosed and over a short period there was a souffle audible at all the valves of the heart, while the heart itself showed some enlargement to the left.

In December, 1944, he suddenly became unconscious. He was very dyspneic and cyanotic, and at the same time coughed up some blood, but in five minutes the condition had improved, although a partial paralysis of the left foot remained.

August, 1945, clubbing of fingers and toes was still obvious. At the back of the neck there was a spot the size of a quarter with telangiectases. The patient had at one time coughed up 300 c.c. of blood. There was no souffle to be heard at the heart or elsewhere in the thorax; the electrocardiogram showed a regular action with a conduction time of 0.15 ventricle 0.07 and negative T_3; no preponderance of right or left. There was also a distinct polycythemia (6,040,000 red blood cells). The circulation time after injection of 2 c.c. magnesium sulfate was 14 seconds.

On the x-ray film of the chest, the same shadow was still visible, now distinctly connected with the hilum by a broad bandlike shadow. Since the shadow on the x-ray film remained unchanged in size and form, the diagnosis of tuberculosis was given up and although at first there had been no signs or symptoms of heart disease the patient was then for several years suspected of having congenital heart disease. However, after some time this diagnosis was reviewed, when we thought that the patient had a cavernous angioma of the lung. This diagnosis was proved correct at operation.

Operation was done April 8, 1947, under endotracheal closed ether anesthesia.* After removal of the seventh right rib, the thorax was opened and in the lower part of the upper lobe of the lung a large number of dark blue tumors could be seen. They projected above the surface of the lung like domes and some of them were as large as a cherry. These tumors were round, very soft on palpation, and pulsated slightly with each heartbeat. They diminished a little in size during inspiration. On compression the tumors disappeared. A very slight thrill could be felt on direct palpation of this part of the lung. The diagnosis of cavernous hemangioma proved to be correct. It was then assumed that there was a very large shunt between arterial and venous systems in the lung and this was proved by the very marked dilatation of the pulmonary artery and vein of this lobe. The symptoms of the patient before operation could all be explained by these findings.

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*Operation by Dr. Boerema, anesthesia by Mrs. Vermeulen-Cranch.

The size of the spongelike tumor was that of a small apple; it was deeply situated in the tissue of the lung, but did not penetrate into the middle lobe. There was only a very shallow cleft between the upper and middle lobes and in this cleft, on the surface of the upper lobe, many large, dark blue, cavernous tumors could be seen. In view of the possibility of a serious hemorrhage it was decided not to divide these two lobes but to remove

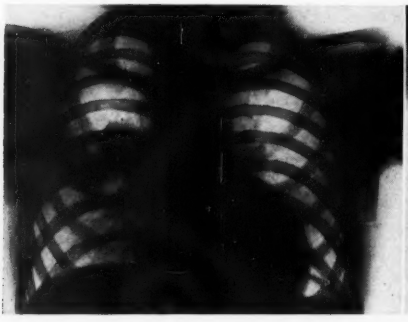

Fig. 1.

Fig. 2.

Fig. 3.

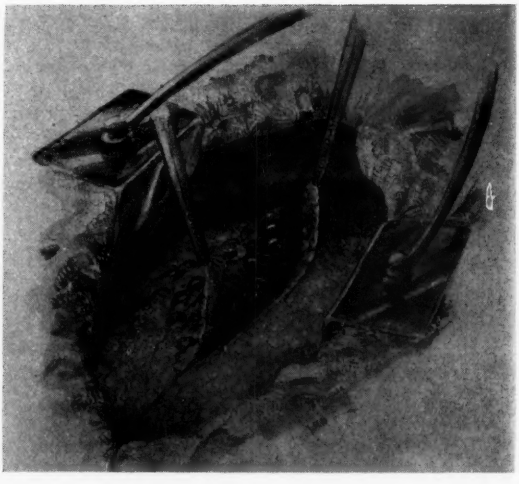

Fig. 4.

the upper lobe together with the middle lobe; a local excision seemed to be impossible. The vessels of the middle lobe were normal, whereas those of the upper lobe were much distended. The bronchus stumps were closed with a single row of end-standing nylon sutures.

The patient made a rapid recovery, showed no more cyanosis, and left the hospital fourteen days after operation. After a few weeks, for the first time in his life he was

able to play and swim with his friends. He felt perfectly well, did not expectorate any more blood, and was not dyspneic.

The specimen showed the spongelike part of the lung, the cavernous spaces of which were filled with blood. The dilated vessels were very thin walled. The spongelike tumor as a whole was not sharply separated from the normal lung tissue.

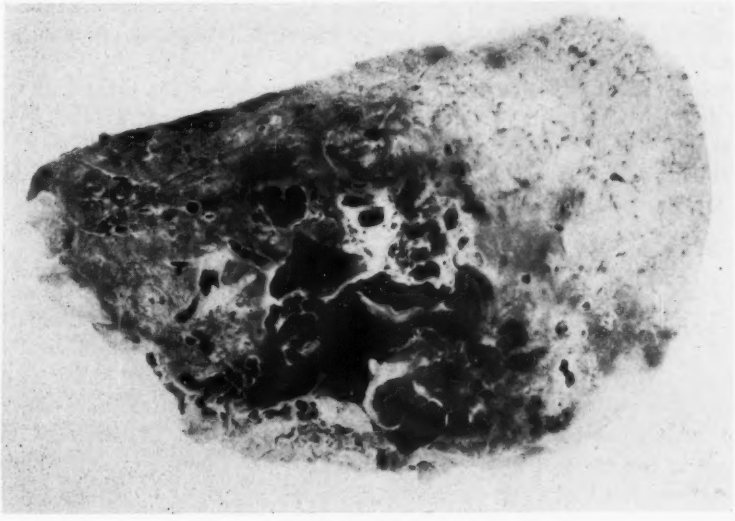

Fig. 5.

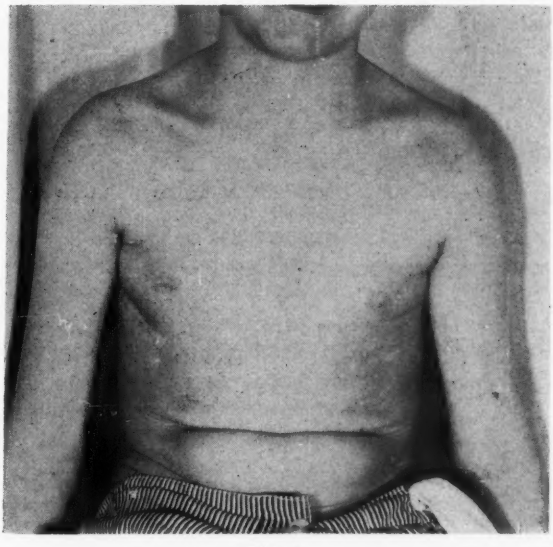

Fig. 6.

Before operation this boy had developed certain complications, any of which might have proved fatal. First, the convulsions in the left side of the body, followed by a partial paralysis of the left foot, may have been caused by thrombosis, as is so often seen in patients with a polycythemia. It is also not improbable that the cerebral symptoms may have been due to the entrance of air into the vessels, at the moment of the rupture of a cavernous space in the lung, particularly in this case, since the convulsions and hemoptysis occurred simultaneously.

Second, during the last few years some dilatation of the heart both to the right and left had occurred. So it is probable that, without operation, a decompensation of the heart would have occurred after some time.

Third, the quantity of blood which was expelled during the hemoptysis was large and it is possible that with a subsequent hemoptysis he might have drowned in his own blood, as recorded in some of the published cases.

It is important to note that in all the published cases of angioma of the lung, other blood vessel tumors, if they were present, were always situated on the head and specially on the lips or at the back of the neck, as in this particular patient. A possible explanation of this may be that since embryologically the lungs are developed from a groove in the ventral wall of the pharynx, or in the head, it seems reasonable to assume that tumors arising from the mesenchyme in this region may be found in both the lung and the head. There were similar findings in the cases of Janes,[1] Mackler and Zion,[2] Lindgren,[3] and Goldman.[4] It is our opinion that these cases strengthen the theory of congenital origin of cavernous angiomas of the lung. The angiomas found in the family of the patient, who has been described by Adams, Thornton, and Eichelberger,[5] were also situated on the head. These latter cases even suggest the probability of the disease originating in the germ cell.

The same theory is sustained also by the cases described by Goldman,[4] which concern two brothers, who both suffered from angioma of the lungs.

So it must be possible to find this disease in very young children. Most of the cases published up to this time concern adults. And as the disease is progressive and after many years shows a dangerous influence on the heart, the angioma of the lung should be operated upon as early as possible.

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ANOXIA DURING INTRATHORACIC OPERATIONS

A Preliminary Report

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THE word anoxia signifies a deficiency in the supply of oxygen available to the cells of the body. The cells which earliest suffer irreparable damage are those of the cerebral cortex, so that in clinical practice it is cerebral anoxia which is significant. A degree of cerebral anoxia sufficient to produce recognizable signs seldom exists after general surgical procedures, but the fact that it can be an important factor in death after major thoracic operations was brought to our attention by three postoperative deaths in a period of two years with clinical and post-mortem evidence of fatal cerebral anoxia.

The signs of severe cerebral anoxia are restlessness, irritability, twitchings, tachycardia, hyperpnea, and rising temperature, leading to coma and death. In a patient who has been subjected to a major operation these may be interpreted as signs of shock, heart failure, or pulmonary atelectasis. At post-mortem the signs are not striking nor characteristic, and may be overlooked. In those patients who survive eighteen hours or more after the onset of the condition there will be cerebral edema.[5] After thirty hours careful histologic study will show changes in the brain,[2] but many patients do not survive that long. It is probable that more deaths have occurred from cerebral anoxia than have been recognized. The picture of "irreversible shock" seen during World War II seems to be identical.

Anoxia is classified as anoxic, anemic, stagnant, or histotoxic. In this investigation we have been concerned with the anoxic type, but it must be borne in mind that during an operation there may be enough blood loss and shock to produce a degree of anemia and stagnation sufficient to contribute appreciably to the total anoxia.

During a thoracotomy the ipsilateral lung is collapsed, and the blood flowing through it, which is not being oxygenated, constitutes a shunt of venous blood to dilute the arterial blood from the functioning contralateral lung. The burden of oxygenation of the blood falls on the contralateral lung and, particularly if it is diseased or has been decreased in volume by a previous lobectomy, anoxic anoxia might be expected to develop. Two of our fatal cases followed lobectomy in patients with bilateral bronchiectasis who had previously had partial resection of the opposite lung.

There is little information available in regard to the degree and duration of anoxia which is likely to be harmful. Investigations carried out in aviation

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medicine have been done on fit individuals, who were not undergoing a surgical operation. These have shown that there is great individual variation in tolerance for anoxia. The anoxic anoxia produced by 14,000 to 18,000 feet of altitude, which produces an oxygen unsaturation of the arterial blood of 16 to 25 per cent, if maintained for an hour will be harmful to the average healthy individual, and very occasionally will be fatal.[6]

The method used in this investigation was to record continuously the level of arterial oxygen unsaturation during and immediately after major thoracic operations by means of an oximeter. This is an instrument developed by G. A. Millikan[4] which, by means of color filters and a two-channel photocell, gives a continuous recording of the oxygen unsaturation in the blood of the fully dilated capillaries of the ear. Many calibrations against direct determinations of arterial oxygen content are reported in the literature to confirm that the instrument is reliable,[3] and in the present investigation the readings have been checked against determinations of the oxygen content of samples of arterial blood taken during operation, as determined by the Van Slyke technique, and it has proved accurate.

Observations have been made in 70 cases, as follows: pneumonectomy, 12; lobectomy, 27; exploratory thoracotomy, 8; other thoracotomies (for example esophagectomy) 8; thoracoplasty, 15. All these operations have been done under intratracheal ether-oxygen anesthesia, with the exception of the thoracoplasties which were done under intratracheal cyclopropane.

In none of the thoracoplasty cases was there any significant fall in oxygen saturation. However, in the other fifty-five cases, in which the pleural space was opened, the oxygen saturation was subject to surprising swings. Invariably, when the bronchi are sucked out there is an abrupt fall to a level of 25 to 30 per cent oxygen unsaturation, followed by a rise over a period of two to three minutes to the mean level for that patient. This rise to the mean level occurs within ten seconds if positive pressure anesthesia up to 15 mm. of mercury is applied. In addition to these violent swings as the result of suction or positive pressure, there is in many patients, as the operation progresses, a gradually increasing degree of oxygen unsaturation commonly to 8 to 18 per cent, which corresponds to an altitude of 15,000 feet, and occasionally from 18 to 30 per cent. Of the 55 thoracotomy cases studied, the first 5 were discarded because of incomplete data, leaving 50 cases. Of these 24, or 48 per cent, showed no oxygen unsaturation; 11, or 22 per cent, showed 4 to 8 per cent; 11, or 22 per cent, showed 8 to 18 per cent; and 3, or 6 per cent, showed over 18 per cent oxygen unsaturation. One patient developed obstruction of the contralateral main bronchus, which could not be relieved; he rapidly developed oxygen unsaturation of over 45 per cent and a few minutes later died of asphyxia.

This gradually increasing oxygen unsaturation can be prevented or corrected by the application of positive pressure anesthesia at a pressure of 15 mm. of mercury. That this phenomenon is not due merely to inflating the nonfunctioning ipsilateral lung is shown by the fact that the rise occurs even after the bronchus on this side is clamped.

When a patient is under general anesthesia there are no significant changes in respiration, pulse, or blood pressure to indicate a serious degree of anoxia, and cyanosis is a late and unreliable sign.[1] Our anesthetists now watch the oximeter and apply positive pressure as indicated. As a result, in the latter part of the series there have been fewer cases showing anoxia.

A further observation which may be of some importance is that at the termination of operation, when the patient is changed over from breathing the anesthetic mixture with its high oxygen content, to breathing air, there is invariably an increase in the oxygen unsaturation of 6 to 10 per cent. A few cases have been followed for several hours after operation and the level has gradually risen to approximately normal over a period of two hours. The use of an oxygen tent has decreased the oxygen unsaturation by only two per cent.

None of the forty-nine patients who survived operation showed any recognizable evidence of cerebral anoxia in the postoperative period.

To explain these observations it is suggested that during intrathoracic operations there is, in about 50 per cent of cases, a gradual diminution in the efficiency of the contralateral lung due to increasing atelectasis associated with a progressive shift of the mediastinum. This lung is re-expanded and its function restored by the application of a positive pressure of 15 mm. of mercury. The sudden profound fall in arterial oxygen saturation associated with bronchial suction is probably due not only to sucking out oxygen, but also to a temporary increase in the atelectasis by negative intrabronchial pressure. Spasm induced by the irritation of the suction catheter may also be a factor.

From this investigation, to date, it is concluded that with this method of anesthesia a significant degree of anoxic anoxia may occur during major intrathoracic operations, that it is probably due to a progressive atelectasis of the contralateral lung, and that it can be prevented or corrected by the use of positive pressure anesthesia. Since anoxia during operation cannot be recognized by clinical signs, the use of the oximeter is a valuable addition to the technique of major intrathoracic operations. The investigation is being continued.

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ANOMALOUS PULMONARY VESSELS

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THE technical advances in thoracic surgery which have made lobectomy and pneumonectomy relatively safe operations have emphasized the clinical importance of anomalies of the vessels supplying the lungs. Prior to these advances in chest surgery, observations on these vessels have been recorded for the most part by anatomists or pathologists. Reference to their occurrence and a classification of the more significant types are lacking in surgical texts and standard anatomic works. Berry,[1] moreover, commented in 1941 on the fact that there is little in the literature regarding the vascular anomalies of the hila of the lungs and urged that they should be reported when encountered. His statement, together with recurring reports of vascular anomalies related to bronchiogenic cysts and experience with three cases of this type, has prompted a review of the problem and the report of these cases with a further case through the courtesy of Dr. Ethan Flagg Butler.

McCotter,[2] in 1910, reported the finding of "an apparently rare case" of a pulmonary artery arising from the thoracic aorta and collected nine cases from the literature. These are summarized in Table I. Batts,[3] in 1939, reported another case and discussed the embryologic aspects. His case, and others reported by nonsurgical workers subsequent to McCotter's review of 1910, are presented in Table II.

Brody,[4] in 1942, reported a case of anomalous pulmonary vein, and summarized 102 such cases previously reported in the literature. Partial drainage of pulmonary veins into the systemic circulation occurred in 68, and of these 74 per cent reached maturity. Compere and Forsyth,[5] discussed this condition and pointed out that the surgeon should be aware of the occurrence of these anomalies.

The first full report of a case of an anomalous pulmonary artery encountered during surgery was that of Harris and Lewis,[6] in 1940. This article also reviewed the comparative anatomy and embryology of accessory pulmonary arteries and veins. Death from hemorrhage occurred in their case. Arce,[7] in 1943, reported a second case in detail. To these reports, four cases are now added. A fatal hemorrhage due to the anomalous vessel occurred in one of these also.

CASE REPORTS

Table III presents the available data on anomalous arteries as reported by surgeons. It includes six instances where brief mention only of the vessel has been made, two reports in personal communications, and the six cases reported in more detail. It would appear that these vessels have been encountered with relative frequency by thoracic surgeons.

Read (by title) at the Twenty-seventh Annual Meeting of The American Association for Thoracic Surgery, St. Louis, Mo., May 28, 29, and 30, 1947.

From the Surgical Service of the Hermann M. Biggs Memorial Hospital, Ithaca, New York.

TABLE I. SUMMARY OF MCCOTTER'S COLLECTED CASES

REPORTED BY	YEAR	AGE	SIDE	ORIGIN	LEVEL	ACCOMPANYING VEIN
			A. *Anomalous vessels*			
Rektorzik	1861	"Girl"	L	Aorta	10 Dorsal	+
Rokitansky	1861	3 mo.	L	Aorta	10 Dorsal	+ (2 arteries)
Ruge	1878	Newborn	L	7th intercostal	–	–
Humphrey	1885	1 yr.	L	Aorta	–	+
Simpson	1907	Term fetus	R	Aorta	10 Dorsal	–
			B. *Anomalous vessels with accessory lobe*			*Comment*
McCotter[2]	1910	65	R	Aorta	7 cm. above Celiac axis, level of 10 Dorsal	7 mm. artery to right lower lobe
Huber	1777	2 yr.	R	Aorta	7 Dorsal	To right lower lobe
Maugars	1802	7 yr.	R & L	Aorta	Below diaphragm	5 mm. artery branches to right and left lower lobes
Meckel	1820	9 mo.	L	Aorta	1 cm. above diaphragm	9 mm. artery to left lower lobe
Hyrtl	1839	Newborn	L	Aorta	Thoracic	Only vessel to left lower lobe

A review of these cases indicates the occurrence of an associated bronchogenic cyst in a significant proportion. We suggested in 1941 that the relation might be of significance. The accumulating evidence indicates that these solitary bronchiogenic cysts are congenital in origin and that they are frequently accompanied by anomalous arteries which are almost entirely congenital in origin also.

The two most significant types of anomalous arteries appear to be (1) those arising from the aorta and passing laterally to the lung. This is the type described by McCotter. It is possible that these are related to the artery described by Turner,[13] which is not infrequently found traversing the pulmonary ligament and giving an arterial supply from aorta to lung. (2) The other group of arteries arise lower down either at or just below the level of the diaphragm. They are of the type described by Batts. These are particularly hazardous from the standpoint of surgical approach.

TABLE II. ANOMALOUS VESSELS—PATHOLOGIC REPORTS

REPORTED BY	YEAR	AGE	SIDE	ORIGIN	COMMENT
Batts, M. J.[3]	1939	40	R	Celiac axis	To right lower lobe
Park[8]	1912	–	R	Right phrenic artery	Base of right lung
Doering[8]	1914	–	R	Innominate artery	Anomalous origin of the right pulmonary artery
Menke[9]	1936	60	R	Right subclavian	Followed right bronchus

It would appear that emphasis should be placed on the possibility of encountering anomalous vessels when surgical extirpation is being carried out of a lesion which falls within the category of bronchiogenic cysts.

CASE 1 (B. M. H. No. 5371).—D. M. D., aged 15 years, female, white, single. The onset of illness was in 1926, when she was treated for "chronic empyema" by open drainage with subsequent healing of the tract. She thereafter remained under observation, roentgenograms showing a persistent rarefied area at the left base; cough continued but was moderate. In 1937 symptoms increased, sputum "tasted bad," and cough was severe.

TABLE III. ANOMALOUS PULMONARY VESSELS

AUTHOR	YEAR	AGE	SEX	ASSOCIATED PATHOLOGY	VESSEL OF ORIGIN	LOCATION
Previous Surgical Reports						
Harris and Lewis[6]	1940	5	F	Abscess L L L	Aorta, thoracic	2 cm. above diaphragm
Moore[10]	1940	–	–	Infected cyst L L	Aorta, thoracic	1 cm. above diaphragm
Berry[1]	1941	–	–	R L L	Aorta, thoracic	Below hilum
Haight[11]	1942	–	–	Cyst L L	?	Below diaphragm
Haight	1942	–	–	Cyst L L	?	Below diaphragm
Arce[7]	1943	15	M	Bronchiectasis L L L	?	Through diaphragm, 2 to 3 cm. outside pericardial attachment to central tendon
Brown and Robbins[12]	1944	21	M	Cyst in acc. lobe L L L	Aorta, thoracic	Above diaphragm
Brown and Robbins[12]	1944	51	M	Cyst, bronchogenic L L L	2 large veins	Through diaphragm
Terrell[14]	1944	48	F	Bronchiectasis L L L (secondary to bronchial adenoma)	Aorta, thoracic	
Bugden[15]	1946	26	F	Unresolved pneumonia L L L with suppuration	Aorta, thoracic	Inferior to inf. pulmonary vein; 4 arteries were 7 mm.; 3 were 3 mm. to 2.5 cm. long
Present Reports						
	1939	15	F	Infected cyst (?) L L L	Aorta	Below diaphragm (?)
	1940	22	F	Abscess of lung L L L	Aorta	Below hilum
	1941	20	F	Congenital cyst L L L	Aorta	Lower border of pulmonary ligament
	1945	13	M	Congenital cyst L L L	Aorta	Between parietal pleura and diaphragm muscle fibers

She was admitted Nov. 4, 1938, x-rays showing "cavity" at the left base posteriorly with fluid level. Open drainage was re-established Nov. 15, 1938, at which time absence of thickened pleura was noted and the correct diagnosis of chronic lung abscess was made. General condition improved but lesion persisted. Lobectomy was therefore done in two stages. On March 30, 1939, the lower lobe was freed and "an anomalous vessel running from either an intercostal artery or the aorta itself was discovered at the lowermost pole. This vessel, approximately 6 mm. in diameter, was isolated but not sectioned at this stage." At Stage 2 on May 9, 1939, the vessel was re-identified. It required suture ligature because of its size. "It contained oxygenated blood—under considerable pressure, and would appear to be an anomalous artery." Lobectomy, completed by mass ligation, was followed by localized empyema and bronchial fistula. Complete healing occurred by July 18, 1939. The patient was discharged, symptomless, on July 29, 1939.

CASE 2 (B. M. H. No. 10399).—C. M.B., aged 22 years, female, white, married. The onset of illness was in 1931, with cough which became productive. Symptoms, relatively mild, recurred at intervals from 1932 to 1938. Sputum then became more copious and late in 1939 became foul. X-ray views on June 17, 1938, showed an ovoid density in the left cardiophrenic angle; on July 7, 1938, fluid level was present.

She was admitted July 11, 1940, roentgenograms showing a sharply outlined area of density 5 by 7 cm. at the left base behind the heart with fluid level present. Lobectomy was done in one stage, July 24, 1940. The left lower lobe was densely adherent in the spinal gutter over an area about 2 cm. in diameter. As the adhesions were cleared, an anomalous vessel was seen projecting from the left wall of the descending aorta to and

into the substance of the left lower lobe. This anomalous vessel was about 1 cm. long between the aorta and the branches which spread into the left lower lobe substance. The diameter was about 4 mm. The vessel was doubly ligated and sectioned. Lobectomy was completed by individual ligation. A localized empyema developed and was drained. Healing occurred October 10. The patient was discharged Oct. 12, 1940, free from cough and expectoration.

The pathologist reported that the anomalous vessel entering the lower part of the lobe could be traced by means of serial cuts—and extended inferior to the main abscess cavity, branching out into finer radicles toward the hilum.

CASE 3 (B. M. H. No. 11184).—M. E. J., aged 20 years, female, white, single. The onset of illness was in January, 1938, with lassitude. Productive cough began August, 1939, followed by twelve pound weight loss. X-ray studies August, 1939, revealed bronchiectasis and possibly pulmonary cyst. Surgical treatment was refused by the patient at this time. Persistent productive cough subsequently caused her to accept operation.

She was admitted Jan. 20, 1941, x-ray views suggesting abscess of left lower lobe with fluid level. Lobectomy in one stage by individual ligation technique was done Feb. 12, 1941. At the very lower end of the pulmonary ligament an anomalous artery approximately 4 mm. in diameter, arising from the descending aorta, entered the substance of the left lower lobe. After this artery had been ligated and sectioned, the left lower lobe could be easily delivered. The postoperative course was uneventful and the patient was discharged on March 14, 1941.

X-ray films were made of the excised lobe before and after instillation of skiodan in the anomalous vessel. The distribution is related to the area of cyst.

The pathologist described the stump of a relatively thick-walled vessel 0.7 cm. in diameter situated medially and posteriorly about 2 cm. above the diaphragmatic border. This vessel was seen on section to travel with branching anteriorly and posteriorly skirting the borders of a large excavated area. A section of the vessel stained for elastic tissue showed a very distinct internal elastic layer which in places presented fine undulations. In the media there was a considerable amount of elastic tissue. The changes in the lung parenchyma strongly suggested congenital cystic disease.

CASE 4 (A. O. M. H. No. 26719).—R. B., male, 13 years of age, white, schoolboy. The patient had had "asthmatic symptoms" since childhood. There had been four attacks of "pneumonia," the first in May, 1938, and the others at approximately two-year intervals. In 1939 the condition was diagnosed "congenital cysts of the left lower lobe" with associated bronchitis and chronic asthma. Previous studies had demonstrated at least fourteen cystic pockets in the left lower lobe, sometimes with, sometimes without fluid levels. Never had there been any evidence of pathologic processes involving the left upper lobe or any portion of the right lung. The boy lived as a semi-invalid because of dyspnea and other "asthmatic symptoms." Nutrition, however, was maintained at a satisfactory level.

The patient had been admitted to the Arnot-Ogden Memorial Hospital (Elmira) from time to time for diagnostic study or medical treatment. He was admitted for definitive surgery on March 25, 1945. Three days later, the general condition being satisfactory, a left lower lobectomy was undertaken. At operation the findings were reported as follows: On opening the left pleural cavity, the left upper lobe was free and quite normal in appearance and texture. The left lower lobe was irregular with ballon-like, bluish, cystic areas interspersed through apparently normal lung tissue. The left lower lobe was everywhere free of adhesions except over the diaphragm and at the lowermost posterior border where it was densely adherent into the sulcus between diaphragm and chest wall, in the spinal gutter. It later developed that the anomalous artery lay at this point. It was about 5 mm. in diameter, and it opened directly from the lowermost thoracic aorta, definitely above the diaphragm but below the pleural reflection. In other respects the hilar structures were normal in their anatomic pattern, and the fissure between the upper and lower lobes was reasonably well developed.

The unusual adhesions at the left base created a problem that required special handling. Inasmuch as the anatomic structures in the pedicle of the lobe were easily accessible, it was decided to carry out all the steps of lobectomy by individual ligation technique and then

attack the basal adhesions when all other structures had been separated. This could be easily done, and finally the lobe was free except for these dense adhesions. The pleural cavity had been entered through the seventh interspace, and the adhesions were at the level of the tenth interspace or lower. It was difficult to obtain free and unobstructed visibility of the adherent area. The dissection was proceeding slowly, partly by blunt dissection, partly by scissors dissection, when there was sudden severe and uncontrollable hemorrhage. In spite of all efforts it was impossible to prevent a fatal outcome.

After the death of the patient it was possible to identify a branch from the aorta arising just below the reflection of the pleura from posterior mediastinum to diaphragm, and just above the fibers of the diaphragm. This branch was about 5 mm. in diameter. It was very short and had obviously been under traction at the time that it had been cut. It must have immediately retracted into the depths of the field, and the stump was too short to grasp.

Comment.—The tragic outcome in this case would probably have been avoided if adequate exposure of the anomalous vessel had been obtained through a lower interspace. This experience emphasizes the necessity of being on guard for congenital vascular anomalies when face to face with even presumptive congenital structural anomalies.

SUMMARY

1. The surgical importance of anomalous pulmonary vessels is apparent.

2. Reports relating to anomalous pulmonary arteries and veins in the anatomic and pathologic literature are tabulated.

3. Four cases encountered during operation are reported.

4. Ten cases previously reported by surgeons are summarized.

5. Two types of anomalous pulmonary arteries are reported by both pathologists and surgeons, the more common type rising from the aorta below the hilum, the second arising from the abdominal aorta or celiac axis.

6. In the pathologists' material anomalies are approximately equally distributed between the right and left sides.

7. In the surgical cases the vessel was on the left side in ten, on the right in one, and the side was not specified in three. The associated pulmonary pathology was an infected cyst in eight instances, a fact which suggests that multiple congenital anomalies were present.

The author wishes to express his indebtedness to Dr. E. F. Butler for his interest and stimulating suggestions, since the first case was encountered on his service in Ithaca in 1939.

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ECTOPIA CORDIS

Report of a Case With Attempted Operative Correction

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THE term "ectopia cordis" has been used to describe all anomalies in which the heart is not within the thoracic cage. Although the congenital defect is a rare one I have found 141 cases in the literature, probably because the anomaly is such a startling one that most of the cases that have occurred have been reported. As far as I have been able to determine only five attempts have been made at surgical correction of this lesion and it is for this reason that this case report is made.

The first description of the anomaly is variously ascribed to Haller or to Martinez, both reporting cases in 1706. Weese, in 1818, was apparently the first to attempt an anatomic classification of the lesion and used the terms (1) ectopia cordis cum sterni fissura, (2) ectopia cordis suprathoracica, and (3) ectopia cordis subthoracica. Townsend, in 1833, preferred the terms (1) cervical heart, (2) pectoral heart with fissure of the sternum, and (3) abdominal heart with a defect in the diaphragm. Perhaps a fourth type, the combined thoracic and abdominal, should be added.

Blatt and Zeldes[1] reviewed the literature in 1942 and found reports of 52 cases, occurring between 1706 and 1938. Roth,[2] in a collective review published in 1939, was able to find record of 83 cases in addition to those listed by Blatt and Zeldes. In addition to the case reported here, I found five additional case reports, making a total of 142 cases.

The location of the ectopia was, in 9 cases collected by Roth,[2] not accurately described so that exact classification was not possible. Of the remaining 133 cases, the site of the defect is shown in Table II.

Of the 52 cases collected by Blatt and Zeldes,[1] 19 infants were premature, 16 were full term, and in 17 the period of gestation was not reported. Of these, 11 were stillborn, 13 lived less than twenty-four hours, 11 lived one to seven days, 3 lived eight to thirty days, 3 lived one to two months, and 8 lived one year or more. Duration of life in 3 was not reported. The life expectancy varies considerably with the site of the ectopia and with the presence of associated abnormalities. Many possessed no covering for the heart, the myocardium being exposed, as in the case here reported; in others a pericardial sac was present and in some skin was also present over the defect in the thoracic wall. In several of the abdominal ectopias no defect was visible.

The prognosis is poorest in the cervical site. Of two cases reported one infant was stillborn and one lived a few hours. Of 30 with thoracic ectopia 5 were stillborn. The remainder lived from one hour to sixteen days. One lived

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TABLE I. CASES OF ECTOPIA CORDIS REPORTED

Author	Number
1. Collected by Blatt and Zeldes	53 (with case report)
2. Collected by Roth (excluding 33 cases also reported by above)	83
3. Hofmann (1938)	
Puddu & Cammarella (1938)	
Kuhnel (1940)	
Sosyal (1940)	
George (1945)	5
4. Present case report	1
Total	142

thirteen months (Greig).[8] The longest duration of life in the infants with combined thoracoabdominal hearts was four months. With the exception of Greig's case all of the infants who survived more than one year had abdominal hearts. The most striking case was that reported by Deschamps who described an autopsy on an old soldier who had died of suppurative nephritis. The left kidney was found to be absent and in its place was the heart within its pericardial sac. The vessels passed through a small hole in the diaphragm. Epigastric ectopia also is not incompatible with life as a number of case reports describe patients in whom an epigastric heart could be grasped in the hand. Two patients were seen when pregnant. One patient was said to be apparently well at the age of 75 years.

Many of the cases had associated congenital anomalies. As might be expected, most of these associated anomalies were cardiac. Greig, in reporting 38 cases from the literature, stated that 2 patients had trilocular hearts, 3 had bilocular hearts, 6 had two superior venae cavae, and 4 had multiple anomalies.

In those cases in which electrocardiographic tracings have been obtained the findings have not been striking. Lintgen[9] reported only slight variation in rhythm. Puddu and Cammarella[4] carried out extensive studies to determine the direction and speed of the electrical impulses at various points on the atria and ventricles. These studies served to corroborate the accepted findings in animal experimentation and confirmed the studies of Barker, Macleod, and Alexander.[13] Barlow[10] felt that the studies in his case tended to disprove the blood vessel theory of conductivity but his tracings revealed some inconsistency. In making tracings with the conventional leads no deflections were noted, indicating that there was no difference in potential between the two halves of the body. When, however, tracings were obtained from electrodes on the surface of the heart and the extremities there was a much greater deflection with the electrode on the right leg than when it was placed on the left. This is difficult to under-

TABLE II. DISTRIBUTION BY SITE OF DEFECT

Site	Number
Thoracic	81
Abdominal	38
Combined thoracoabdominal	10
Cervical	4
Unclassified	9
Total	142

Fig. 1.

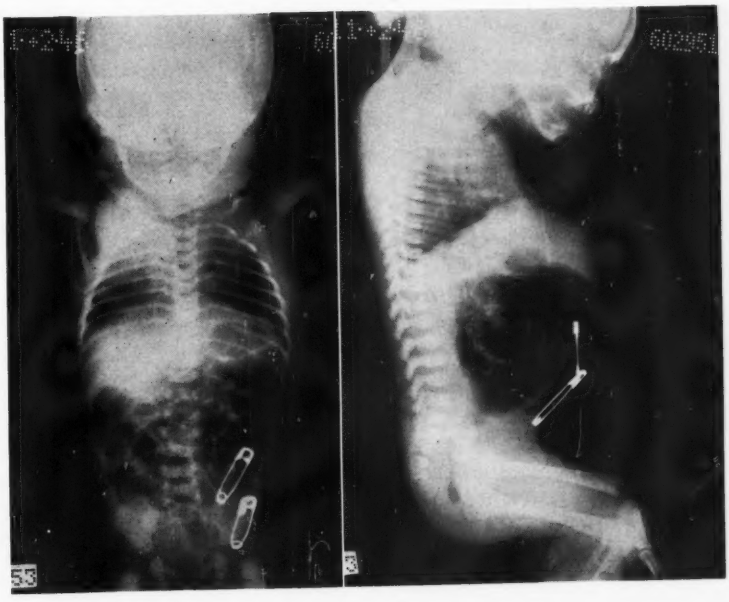

Fig. 2.

Fig. 3.

stand in view of the failure to obtain deflections in the conventional leads which would indicate a lack of potential between the two extremities. The tracings obtained by Blatt and Zeldes were not strikingly abnormal. In our case the findings were without value as they were taken with a moist saline pad on the heart.

CASE REPORT

Baby Girl S. was referred to the University of Michigan Hospital for surgical correction of an external heart and was admitted eighteen hours after delivery. The period of gestation had been normal with the exception of hyperemesis gravidarum during the first six months. The mother had had no previous pregnancies. The delivery was described as normal. Upon admission the temperature was 98.4 degrees, the hemoglobin 15.4 Gm., and the leucocyte count 19,600.

Fig. 4.—The heavy line represents the incision used to form a skin flap and gain access to the left pleural cavity.

The physical examination was not remarkable save for the evident anomalies. The manubrium was present but the sternum was totally absent. The heart was completely external and uncovered by any membrane, the myocardium being exposed. It was vertical in position with the apex, which was bifid, pointing straight up and, at times, beating against the infant's chin. The heart surface dried readily and had to be kept moistened. The blood flow through the coronary artery, which was single, the right coronary artery being absent, could readily be seen. The atria consisted of short cylindrical appendages which shortened upon contraction but decreased little in caliber. The right ventricle seemed larger than the left. The great vessels entered the heart through a 2.5 cm. opening just inferior to the lower border of the manubrium. The vena cavae, the aorta, and the pulmonary artery could be distinguished. The skin below this opening was absent and in its place was an irregular area of grayish gelatinous-appearing tissue which extended down to the umbilicus. This area had roughly the shape of the heart when it was placed against the body. The prominence of the

upper abdomen was striking and it appeared that this prominence was due to the protrusion of the liver beneath the weakened abdominal wall.

A motion picture, in color, of the functioning heart was obtained. The x-ray views revealed the absence of an intrathoracic heart.

It was explained to the child's father that operation offered little chance of correcting the defect but he was extremely anxious to have an attempt made. From an operative standpoint a number of difficulties presented themselves. First, the absence of normal skin inferior to the heart made placement of an incision difficult; second, it was probable that the heart would not tolerate displacement from the vertical position; and third, there seemed to be no place to put the heart.

Under ether anesthesia an incision was made horizontally from the opening in the midline and carried down vertically toward the left nipple. The skin and subcutaneous tissue was then undermined to the common cartilages, giving exposure almost to the midline. The pleura was thus exposed and the absence of a pericardial sac verified. The only way to place the heart beneath the skin flap was to place it within the left pleural cavity and this was done. However, the cardiac beat became very weak when this maneuver was carried out and accordingly it was returned to its original position. Another attempt was made but again the beat became feeble. Since further procedure seemed fruitless the chest was closed under slight positive pressure and the skin resutured. At the conclusion of the operation the cardiac beat was exceedingly feeble and failed to respond to the intracardiac injection of adrenalin. Death occurred approximately twenty-two hours after birth.

The post-mortem examination revealed a tetralogy of Fallot. There was hypoplasia of the pulmonary conus and aplasia of the root of the pulmonary artery, a widely patent ductus arteriosus, an interventricular septal defect, and a large patent foramen ovale. Only the left coronary artery was present. There were also an incomplete descent of the cecum and abdominal ascites.

DISCUSSION

Previous attempts at operative correction have been made by Cutler and Wilens,[11] Bloch,[12] and Hofmann.[3] I have learned that E. Finocchietto also attempted the operation but did not report the case. In Bloch's case the child lived seven days after the heart was placed beneath skin flaps but its action was impaired and the sutures had to be removed. In Hofmann's case the baby died during an attempt to place the heart beneath the skin. Cutler and Wilens made an oblique incision downward from the ring and exposed the pleura. As in my case, the prominence of the liver prevented the creation of an adequate pocket for reception of the heart. When the defect was nearly closed by approximating the skin flaps, the heart was throttled by the tension. Recognizing the futility of further attempts they removed the sutures. The child lived eleven hours after operation. The operative findings in each case have been similar and it would appear the corrective attempts are not justified.

The absence of normal skin below the defect in the chest has been present in many of the cases. The best explanation of this is put forth by Barlow. Since, in this case, this skin defect was roughly the shape of the heart when it was forced against the chest wall he felt that the constant pressure and irritation of the beating heart during uterine life was the cause.

SUMMARY

A case of ectopia cordis with an attempt at operative correction is presented. On the basis of the operative findings in this case and in the four other cases

subjected to operation by other authors, correction of the defect does not appear to be feasible.

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NOTICE

The twenty-ninth annual meeting of the American Association for Thoracic Surgery will be held on March 29, 30, and 31, 1949, in New Orleans, La. Headquarters will be at the Roosevelt Hotel.

Requests for hotel reservations should be mailed direct to Dr. Alton Ochsner, 1430 Tulane Avenue, New Orleans 13, La.

Abstracts for presentation of papers for the 1949 meeting must be received for consideration by the Program Committee, on or before Jan. 1, 1949. These abstracts should be submitted in *quadruplicate* and addressed to the Secretary of the Association, Dr. Brian Blades, George Washington University Hospital, 901 23rd St., N.W., Washington 7, D. C. They should not be sent to the Editor. The early date of the 1949 meeting makes it absolutely necessary that these abstracts be received before the deadline date. Abstracts received after Jan. 1, 1949, will not be considered.

Manuscripts for competition in the Rose Lampert Graff Prize should be sent directly to the Secretary of the Association not later than Jan. 1, 1949.